Predisposing and maintaining factors in OCD and hoarding disorder

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Heritability of hoarding symptoms: a systematic review of twin studies

SYSTEMATIC REVIEW

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1. ABSTRACT

Background: The causes of Hoarding Disorder, a newly recognised psychiatric disorder, are unknown. A number of recent twin studies have suggested that hoarding symptoms are heritable but heritability estimates vary across studies and the reasons for this remain unclear. Findings from two recent twin studies have suggested a dynamic picture with age- and gender-specific risk factors accounting for the variation across studies.

Aim: The present systematic review aims to provide the first, comprehensive, and up-to-date review of twin studies of hoarding symptoms, with a view of clarifying and shedding light on gender- and age-related changes in heritability for HD.

Methods: PubMed, PsycINFO, Medline, Embase, and Web of Science were searched up to March 2016 using relevant key search and MeSH terms, according to PRISMA guidelines. The quality of studies was assessed using a revised 11-items checklist for cross-sectional/prevalence studies assessing the three major domains of risk of bias.

Results: a total of six studies met inclusion criteria. The methodological quality of included studies was moderate-to-high for selection and methodological bias, but overall poor for confounding bias. Genetic factors play an important role in the aetiology of hoarding symptoms across all studies. Genetic factors seem to play a stable and significant role for male hoarding behaviours. For women, on the other hand, these influences appear to vary across development, with shared environmental factors predisposing young females to hoarding symptoms and genes playing a more influential role only later in life.

Conclusions: hoarding symptoms are moderately heritable; the extent of genetic influences on hoarding however is likely to change during development and differ between genders. The current review supports genetic research and further examination of environmental factors predisposing individuals to hoarding symptoms. More research, including longitudinal twin studies, is needed to conclusively identify and compare risk factors for hoarding across genders and age groups.

2. INTRODUCTION

Hoarding Disorder (HD) is a mental disorder than has been recently included in the *Obsessive Compulsive and Related Disorders (OCRD)* chapter in DSM-5, alongside Obsessive Compulsive Disorder (OCD), Body Dysmorphic Disorder (BDD), Trichotillomania (TTM), and Skin Picking Disorder (SPD). This condition is defined as a persistent difficulty discarding or parting with possessions, resulting in clutter and causing clinically significant distress and/or functional impairment (American Psychiatric Association, 2013). The DSM-5 diagnostic criteria for HD also include additional items to specify the extent of 'excessive acquisition' and to denote the level of insight in relation to the patient's hoarding behaviour (see *Appendix 1* for full diagnostic criteria). OCRDs, including HD, are thought to be phenomenologically and aetiologically related to Obsessive Compulsive Disorder (OCD) prompting its inclusion in the new chapter in DSM-5 (American Psychiatric Association, 2013) (*Appendix 2*). Despite the similarities across OCRDs, the question as to whether these conditions are related and should be grouped together as separate disorders remains the source of an on-going debate. Of particular note, the evidence for hoarding as a separate diagnosis in DSM-5 has been extensively debated. Historically, hoarding has been considered a symptom of OCD. Research however has shown that most hoarders do not endorse OCD symptoms and that hoarding can manifest itself as a distinct set of non-OCD related symptoms; differences between OCD and hoarding include cognitive-behavioural processes, course of the illness, neurobiological substrates, and treatment response (Mataix-Cols et al., 2010; Pertusa et al., 2008). These observed differences landed support for the creation of a new diagnostic category to describe cases where hoarding occurs in the absence of OCD or any psychiatric disorders, as well as developmental and neurological conditions. The decision for HD to be classified in the OCRDs chapter on the other hand was largely dictated by the fact that most hoarding research has been done in the context of OCD.

A number of important advantages as well as disadvantages have been considered leading up to the inclusion of HD as a separate condition in DSM-V. Among the main advantages of including hoarding as a separate disorder were the potential increase in public awareness, improvement in identification of cases, accuracy of diagnosis, and tailoring of treatment (Mataix-Cols et al., 2010). Although compulsive hoarding comorbids with other conditions, including OCD, it can and often presents in isolation and severe forms to necessitate specific treatment. The creation of a new diagnosis in DSM-V would address much of this unmet need. It would also likely stimulate research into the aetiology and treatment of compulsive hoarding using an agreed-upon set of diagnostic criteria.

The reported disadvantages for its inclusion as a separate disorder included the misuse of a HD diagnosis in a way that would produce harm, such as the pathologizing of normal behaviour. Of note, however, research supporting the current diagnostic criteria for HD has been found to discriminate between adaptive and maladaptive degrees of hoarding behaviour with high reliability. Social and economic consequences of a new disorder with an estimated prevalence between 2 and 5% in the general population have also been discussed as potential disadvantages of including HD as a separate condition in DSM-5, with important financial implications for treatment and therefore for the health systems.
Overall, in spite of the on-going debates, research on HD has highlighted the potential benefits in favour of its inclusion (e.g. improvement in clinical communication, patient care, research) as out weighting the potential harms (e.g. misuse/misdiagnosis, pathologizing normal behaviours).

While HD was initially considered to be a relatively rare condition, recent epidemiological research has now shown that clinically significant hoarding behaviours affect as many as 2-5% of the general population (Iervolino et al., 2009; Mueller et al., 2009; Samuels et al., 2008; Timpano et al., 2011), with approximately 1.5% meeting full diagnostic criteria for HD (Nordsletten et al., 2013). In the largest epidemiological study to date, HD was found to be equally prevalent across genders (Nordsletten et al., 2013). Although some studies have reported higher rates in men (Iervolino et al., 2009; Samuels et al., 2008), rates of women have been found to predominate in other studies, particularly clinical HD and adolescent samples (Ivanon et al., 2013; Lopes-Sola et al., 2014; Samuels et al., 2008; Steketee et al., 2015). As such, it remains unclear whether hoarding affect men and women to the same extent and whether there may be differential etiological influences accounting for gender differences in the prevalence and presentation of HD in some studies. In terms of its onset and course, HD is a chronic condition with a typical onset around early to mid-adolescence and a reported increase in symptoms severity with age (Ayers et al., 2010; Grisham et al., 2006; Ivanov et al., 2013; Kim et al 2001; Seedat & Stein, 2002). Psychiatric comorbidity is also common in HD, with up to 70% of hoarders presenting one other comorbid psychiatric condition, most commonly anxiety and/or depression (Frost et al., 2011).
The etiology of HD remain largely unknown, though likely to be multifactorial in nature and related to a complex interplay of genetic, neurobiological, and psychosocial factors. The evidence for a role of genetic factors in the etiology of HD comes from a multitude of research designs, including family, twin, and molecular genetic studies. Uncontrolled family studies have consistently shown that compulsive hoarding is a familial condition (Frost & Gross, 1993; Pertusa

et al., 2008; Samuels et al., 2002; Samuels et al., 2007a, 2007b; Steketee et al., 2015). Hoarding sufferers (with or without OCD) report a high prevalence of hoarding amongst their relatives, with rates ranging from 49% to 57% (Samuels et al., 2002, 2007; Pertusa et al., 2008; Steketee et al., 2015). Of note, Steketee and colleagues examined the familial pattern of hoarding symptoms by collecting data on family history from 217 adults with clinically significant hoarding (HD), 96 OCD cases, and 130 healthy controls. The authors observed a greater frequency of hoarding amongst female relatives; that is, more mothers and sisters were reported to have hoarding symptoms than fathers and brothers of hoarders. In this study, parents were also more commonly reported to endorse hoarding symptoms in comparisons to siblings; the authors argue this may be due to their older age and hoarding symptoms severity being shown to increase with age. Yet to be explored, it is possible the predominance of hoarding symptoms among females and older relatives (Steketee et al., 2015) indicates sex and age effects in genetic transmission of hoarding behaviours.

A number of recent twin studies have allowed estimation of the proportion to which this observed familiality may be due to genetics (i.e. heritability) versus environmental factors. A handful of twin studies have been carried out since 2009, demonstrating that hoarding is a moderately heritable condition, with genes accounting for up to 50% of the variance in symptoms; the remaining variance was due to non-shared or unique environmental factors and measurement error, with shared environmental factors playing a negligible role in most studies (Iervolino et al., 2009; Iervolino et al., 2011; Ivanov et al., 2013; Lopez-Sola et al., 2014; Mathews et al., 2014; Taylor et al., 2010). Despite evidence for its heritability, estimates of hoarding heritability vary between studies and the reasons for this remain unclear. While some variation is natural due to differences between populations and settings, factors such as age and gender may also explain some of this heterogeneity and variation. Indeed, Ivanov et al (2013) reported significant shared environment influences on hoarding behaviours among adolescent female twins - a finding that was not found for teen male twins and in adult samples - sparking a debate on the importance of shared environment versus a genes in hoarding for females in this age group. It is possible that at least some of the genes influencing hoarding behaviour are different for males and females, potentially accounting for the mixed findings on the prevalence of HD across genders. Similarly, it is possible that genetic and environmental influences on HD change across development, with environmental accounting for relatively more variance in child hoarding behaviour and genetic factors accounting for more variance in adult hoarding behaviour. These developmentally-based

changes in the relative influences of genetic and environmental factors may furthermore differ between genders.

No systematic review on genetics of hoarding symptoms was identified in the Database of abstracts of reviews of effects (DARE), the Cochrane database of systematic reviews (CDSR) and the Database of promoting health effectiveness reviews (DoPHER). The present systematic review aims to provide the first, comprehensive, and up-to-date review of twin studies on hoarding in order to fully examine the role of genetic versus environmental factors predisposing to hoarding symptoms, and with a view of shedding light on any gender- and age-related changes in heritability for hoarding.

2.1. The twin method

Twin studies provide an excellent mean to estimate the proportion to which a trait or disorder is influenced by genes versus environment. Specifically, twin analyses seek to decompose the phenotypic variance into three factors: A (additive genetic or heritability, i.e. the proportion of phenotypic variation that can be attributed to genetic factors), C (common/shared environment, i.e. environmental effects shared by twins) and E (unique/non-shared environment, i.e. environmental effects unique to each twin, plus measurement error). While MZ twins share a 100% of their genetic makeup, DZ twins share on average only half of their segregating genes. The twin model is based on the difference in genetic sharing between MZ and DZ pairs and on the assumption that MZ and DZ twin pairs share the same family environment. Comparing the correlations within MZ and DZ pairs therefore provides a very first impression of the contribution of genetic versus environmental factors on a phenotype or trait of interest. For instance, greater MZ versus DZ correlations suggest a genetic contribution to phenotypic variation. An MZ correlation less than 1.0 on the other hand is indicative of significant unique environmental factors. Model-fitting analysis then utilizes this information to quantify and separate the observed phenotypic variance into A, C, and E.

Structure Equation Modelling (SEM) is currently the main analytical approach to twin data. Specifically, in twin analyses, MZ/DZ correlations are first estimated. Maximum-likelihood model-fitting analyses (Neale and Cardon, 1992) are then undertaken to estimate the contribution of genetic and environmental factors on the phenotype of interest, decomposing its variance into A, C, and E components. To this end, data is fitted to a saturated model, in which twin correlations

are estimated freely without any constraints. Goodness of fit is assessed by comparing the -2 log-likelihood chi-square values of the saturated to an ACE model. In studies including male and female twins, sex-limitation models can also be fitted to the raw data to test for *qualitative* and *quantitative* sex differences. *Qualitative* sex differences assume distinct genetic and/or environmental factors for males and females and are typically implied when correlations for DZ opposite sex (DZOS) twins are significantly less than same-sex DZ twin correlations. *Quantitative* sex differences on the other hand refer to variations in the magnitude of genetic and/or environmental influences across genders, so that sex differences are mainly quantitative; these differences are typically suspected when male DZ correlations differ from female DZ twin correlations. Finally, to explain the observed data and pattern of variance using as few parameters as possible, reduced submodels, where the genetic parameter (CE model), shared environmental parameter (AE model), and both these parameters are dropped (E model), are tested and compared to the full ACE model. The difference in the chi-square value relative to the change in degrees of freedom provides an indication of the goodness of fit and parsimony, allowing to select the most parsimonious model that fits the data best (i.e. the best fitting model) (Neale and Cardon, 1992).

3. METHODS

3.1. Literature Search

The literature search was completed following PRISMA guidelines (Moher et al., 2009). Specifically, a search of the PubMed, Embase, Medline, and PsycINFO databases was carried out using relevant search terms related to hoarding and genetics (see *Appendix 2* for key search terms). The initial search was completed in March 2016 without any restrictions or filters. After removal of duplicates, records were screened at the title and/or abstract levels. The full-text of the relevant papers was assessed for inclusion/exclusion and the references of identified articles also reviewed.

3.2. Inclusion/Exclusion Criteria

Given the overall paucity of research on HD, we aimed to include as many studies as possible. As such, any twin study reporting on heritability estimates on the hoarding phenotype regardless of study design (e.g. volunteer, population-based or register-based) was considered eligible for inclusion in the current review. Records were excluded if they were not a twin study (e.g. family, molecular genetic, animal studies) or did not report heritability estimates for the phenotype of interest. Twin studies on the same samples were included as long as the instrument utilised differed between studies (e.g. Iervolino et al., 2009 and Iervolino et al., 2011); on the other hand, when several publications reported on the same twin sample and measures, we opted to include the largest one reporting on univariate twin analyses on hoarding symptoms.

3.3. Quality Assessment

Two recent systematic reviews compiled a list of quality assessment tools for various study designs, emphasising the lack of such tools for genetic studies (Zeng et al., 2014; Sanderson et al., 2007). The quality of the included twin studies in the current review was therefore assessed using a revised checklist for cross-sectional/ prevalence studies, which was developed according to the Agency for Healthcare Research and Quality (AHRD; http://www.ncbi.nlm.gov/books/NBK35156/) criteria. The choice of this quality assessment tool was largely based on its use in previous systematic reviews of twin studies (e.g. Wang et al., 2015), recommendations from the Strengthening the Reporting of Observational Studies in

Epidemiological Statement (STROBE; Von Elm et al., 2007), and its validity according to a recent systematic review on quality assessment tools (Zeng et al., 2014).

The 11-items checklist was modified strictly following guidelines from a recent systematic review of twin studies (e.g. Wang et al., 2015). As such, three items (items 4, 5, and 11) were removed; the revised checklist included eight criteria assessing the three fundamental domains of risk of bias, namely selection (source of information, inclusion/exclusion criteria, time period used for identifying twins, missing data, and response rate), measurement (quality assurance), and confounding bias (describe how confounding was assessed and/or controlled) (Sanderson et al., 2007; Wang et al., 2015).

3.4. Data Extraction

For each included study, data were extracted on publication information (author and publication year), study' characteristics (country, source, sample size, age, and gender), methods (zygosity determinant, hoarding measure, and twin analysis) and relevant results (twin correlations and heritability estimates, including 95% CI when available) (*Appendix 3* for Data Extraction Form).

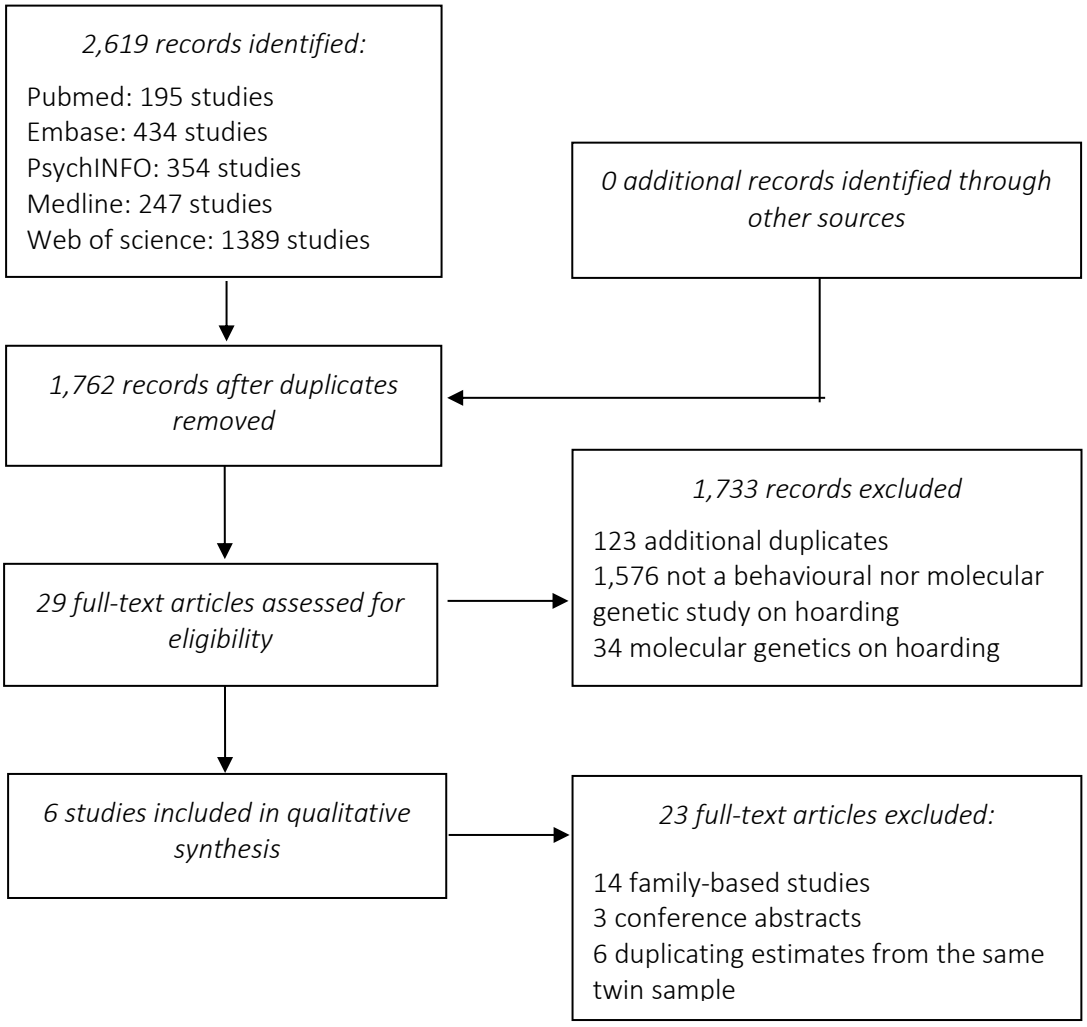
The literature search and quality assessment were completed independently by two trained researchers with any disagreements/discrepancies being resolved through discussion and consultation with other co-authors.

4. RESULTS

4.1. Literature search

The literature search identified 2,619 records/hits from PubMed, Embase, Medline, and PsychInfo using the aforementioned keywords (*Figure 1*). After removal of duplicated (N=857), 1,762 records were screened at the title/abstract level; 29 full-text articles on behavioural genetics were selected as potentially eligible, 6 of which met our inclusion criteria and were included in the review (Iervolino et al., 2009; Iervolino et al., 2011; Ivanov et al., 2013; Lopez-Sola et al., 2014; Taylor et al., 2010; Mathews et al., 2014); no additional articles were identified via inspection of the reference of selected articles. *Table 1* provides a summary on the included twin studies.

Figure 1. PRISMA (2009) Flow Diagram



4.2. Profile of the included studies

A total of six independent twin studies on hoarding were completed between 2009-2016 across five countries (UK, Sweden, Australia, Netherlands, and Canada).

Most of these studies (N=5; 83%) were carried out using voluntary twin registries (Iervolino et al., 2009; Iervolino et al., 2011; Ivanov et al., 2013; Lopez-Sola et al., 2014; Mathews et al., 2014), with the exception of one study where twins were recruited from the community via advertisements (Taylor et al., 2010).

Sample sizes ranged from 614 (Taylor et al., 2010) to 7,906 (Mathews et al., 2014), with a mean age of 15 to 55.5 years and age ranges of 15 to 97 years. Though predominantly females (78.1%), four of the six studies included both male and female twin pairs (Ivanov et al., 2013; Lopez-Sola et al., 2014; Mathews et al., 2014; Taylor et al., 2010); two other studies carried out twin analyses on female twins only as the small number of male respondents did not allow sufficient power to investigate quantitative and qualitative sex differences in the heritability of hoarding symptoms.

Zygosity determination was established via both questionnaires and DNA (fingerprints or genome-wide scans) (Iervolino et al., 2009; Iervolino et al., 2011) or via DNA testing only (Ivanov et al., 2013). The questionnaire method was used as the only source for zygosity determination in the other three studies (Lopez-Sola et al., 2014; Mathews et al., 2014; Taylor et al., 2010), with a reported accuracy rate of 95% compared to DNA testing.

All studies utilised data obtained via self-report questionnaires: two studies (33%) used the Obsessive-Compulsive Inventory-Revised (OCI-R) (Foa et al., 2002) to assess hoarding symptoms and its heritability (Iervolino et al., 2011; Taylor et al., 2010). The OCI-R is an 18-items, 5-point Likert scale questionnaire designed to assess the distress associated with various OCD symptoms; it includes three items which measure the severity of hoarding symptoms with total subscale scores ranging from 0-12. The Hoarding Rating Scale- Self Reported (HRS-SR) (Tolin et al., 2010) was employed in the remaining four studies (67%) (Iervolino et al., 2009; Ivanov et al., 2013; Lopez-Sola et al., 2014; Mathews et al., 2014), one of which was a twin study of hoarding in adolescence. The HRS-SR is a 5-item self-report questionnaire measuring clutter, difficulty discarding, excessive acquisition, distress, and impairment on an 9-point Likert scale. Strong psychometric properties have been reported for both scales (Abramowits et al., 2006; Foa et al., 2002; Tolin et al., 2008a; Tolin et al., 2008b; Tolin et al., 2010), though the use of the HRS-SR in an adolescent population has yet to examined and validated.

Finally, Log transformation of twin data or *liability-threshold modelling* are statistical strategies applied in cases of non-normal data distribution or categorical data. Due to data skewness in all studies, the majority of studies used Liability threshold modelling to estimate the contribution of genetic and environmental influences on compulsive hoarding (Iervolino et al., 2009; Iervolino et al 2011; Mathews et al., 2014; Taylor et al., 2010); two studies on the other hand used SEM on transformed continuous data instead (Ivanov et al., 2013; Lopez-Sola et al., 2014).

Table 1. Details of twin studies on hoarding included in the review

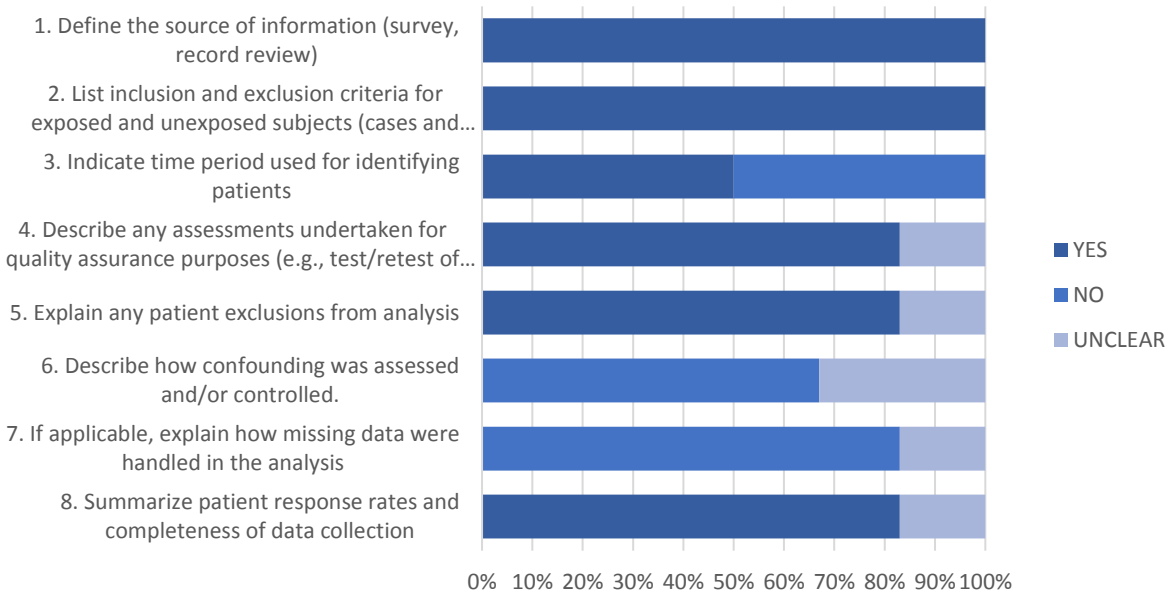
Author	Year	Country	Source	N	Mean age (range)	Sex (% females)	Zygosity determinant	Instrument	Methods
Iervolino et al	2009	UK	UK adult twin registry	4,355	55.5 (17-86)	100%	Questionnaire + DNA	HRS-SR	SEM LTM
Taylor et al	2010	Canada	Community sample	614	40.0 (17-81)	78%	Questionnaire	OCI-R	SEM LTM
Iervolino et al	2011	UK	UK adult twin registry	4,355	55.5 (17-86)	100%	Questionnaire + DNA	OCI-R	SEM LTM
Ivanov et al	2013	Sweden	Sweden twin registry	3,110	15	55%	Questionnaire + DNA	HRS-SR	SEM on transformed continuous data
Lopez-Sola et al	2014	Australia	Australian twin registry	2,495	34.2 (18-45)	59%	Questionnaire	HRS-SR	SEM on transformed continuous data
Mathews et al	2014	Netherlands	Netherlands twin registry	7,906	33.2 (17-97)	69%	Questionnaire	HRS-SR	SEM LTM

Abbreviations: HRS-SR, Hoarding Rating Scale- Self-Repot; OCI-R, Obsessive Compulsive Inventory-Revised; SEM, Structural Equation Modeling; LTM, Liability Threshold Modelling.

4.3. Methodological quality of the included studies

An overview of the methodological quality assessment of included studies is provided in *Figure 2*. All studies (100%) defined their sample appropriately, by defining the source of information and inclusion/exclusion criteria (*Item 1* and *2*). Most studies (83%) provided an exhaustive summary concerning quality assurance (*Item 4*) and response rate and completeness of data collection (*Item 8*); indeed, two studies (Lopez-Sola et al., 2014; Taylor et al., 2010) failed to provide clarification or details regarding quality assurance and response rate. Whilst all studies provided references for further information on the twin sample (e.g. in previous studies completed by the research group or twin registry website), only half of the studies explicitly stated information on excluded twins (*Item 5*) and the time frame used for identifying patients (*Item 3*) (Ivanov et al., 2013; Lopez-Sola et al., 2014; Mathews et al., 2014). Finally, adjustment for confounders (*Item* 6) and how missing data was handled (*Item 7*) was either not reported or unclear from the published articles. The low percentage of description on how missing data and confounding variables were assessed and controlled for in the analysis might underscore a potential for selection and confounding bias in most twin studies. A detailed summary of the quality assessment of each study can be found in the Appendix 5.

Figure 2. Methodological quality assessment



4.4. Twin correlations

A total of five out of six studies reported MZ and DZ twin correlations as shown in *Table 2.* Irrespective of gender, overall, most studies report a higher MZ versus DZ twin correlation, indicative of genetic factors playing a role in predisposing individuals to hoarding; the moderate MZ correlations however are also indicative of unique environmental influences. Taking gender into account, similar patterns of twin correlations (i.e. strongest in MZ than in DZ pairs) were observed for male twins across all studies (Ivanov et al., 2013; Mathews et al., 2014; Lopez-Sola et al., 2014), albeit with a potential, slight decrease in MZ correlations in studies using older samples. This finding suggests a meaningful genetic contribution to hoarding in males, with potential heritability changes from adolescence to adulthood. A different pattern of results was observed for female twins; no significant differences were found in MZ versus DZ correlations for adolescent girls (Ivanov et al., 2013), suggesting similarities between female twins could be explained by shared environmental factors rather than genes at this age. A higher, yet moderate, MZ versus DZ correlation pattern is observed again in older female twin pairs (Mathews et al., 2014; Lopez-Sola et al., 2014; Iervolino et al., 2009; Iervolino et al., 2011), indicating genetic and unique environmental factors at play in older female twins. Finally, DZ correlations for boys were significantly lower than DZ correlations in girls in Ivanov et al (2013)' study. DZOS correlations were overall lower than same-sex correlations; this difference was particularly evident among adolescents (Ivanov et al., 2013). In older twins, the resemblance between DZOS twins seem to decrease over time. Overall, results on DZ correlations (i.e. DZM<DZF in adolescence, same-sex DZ > DZOS, and DZOS correlation decreasing over time) is indicative of quantitative and qualitative sex differences across different age groups.

Table 2. Monozygotic and Dizygostic twin correlations (95% CI) for each study, arranged by samples' mean age

	MZf	DZf	MZm	DZm	DZOS
Ivanov et al., 2013	0.35 (.25-.44)	0.41 (.31-.51)	0.44 (.33-.54)	0.17 (.06-.29)	0.16 (.07-.25)
Mathews et al., 2014	0.34	0.17	0.36	0.18	0.09
Lopez-Sola et al., 2014	0.39 (.29-.48)	0.19 (.05-.32)	0.25 (.11-.37)	0.14 (-.05-.31)	0.10 (-.07-.27)
Taylor et al., 2010	n.r.	n.r.	n.r.	n.r.	n.r.
Iervolino et al 2009	0.52 (.45-.57)	0.27 (.19-35)	n.a.	n.a.	n.a.
Iervolino et al 2011	0.50	0.27	n.a.	n.a.	n.a.

Abbreviations: MZ, monozygotic; DZ, dizygotic; DZOS, dizygotic opposite sex; f, females; m, males; n.a., not applicable; n.r. not reported

4.5. Genetic and environmental influences on hoarding

Table 3 and *Figure 3* report estimates of genetic and environmental influences on hoarding symptoms for each study.

The AE model was reported as the best fitting model in all twin studies except for female adolescent twins in Ivanov et al (2013), where shared environmental factors were found to play a significant role (but not male twins). Three studies (Iervolino et al., 2009, Iervolino et al., 2011, Ivanov et al., 2013) provided estimates for all three parameters (i.e. A, C, and E), whilst three others (Lopez-Sola et al., 2014; Mathews et al., 2014; Taylor et al., 2010) only reported the best fitting AE model estimates.

With regards to sex effects, four studies examined gender differences in heritability of hoarding. No *qualitative* sex differences were observed in any of the included studies. Two studies on the other hand found *quantitative* sex effects, that is, a difference in the magnitude of genetic and environmental influences on hoarding symptoms between sexes (Ivanov et al., 2013; Lopez-Sola et al., 2014). Specifically, greater heritability estimates were reported for adolescent boys (32%) compared to girls (2%) in Ivanov et al (2013); the opposite trend was observed in a young adults (Lopez-Sola et al., 2014), where authors reported a tendency towards greater heritability of hoarding in females (38%) versus male (25%) twins. In contrast, Mathews et al (2014) and Taylor et al (2010) did not find sex differences in heritability of hoarding symptoms, though in the later study, sex-limitation models were not fitted and tested against the best-fitting AE model as per standard analytical procedures to model such effects.

Overall, hoarding symptoms were moderately heritable, with estimates ranging from 2% (Ivanon et al., 2013) to 49% (Iervolino et al., 2009). For males, heritability estimates were 32% (Ivanov et al., 2013) and between 25%- 34% (Lopez-Sola et al., 2014; Mathews et al., 2014) for adolescent and adult males, respectively; as shown in *Table 3*, the 95% CI for heritability estimates for males across the different studies and age samples largely overlap, providing no strong evidence in favour of change in heritability for men across development so far. More variation in results were observed among female samples. Genetic factors played a negligible role among adolescent girls (2%; 95% CI 0-24) (Ivanov et al., 2013), whilst greater heritability was reported in studies on older female twin samples, estimated at 34% (95% CI 15-53%) (Mathews et al., 2014), 38% (95% CI 29-47) (Lopez-Sola et al., 2014), 44% (95% CI 29-54) (Iervolino et al., 2011), and 49% (95% CI 30-57)

(Iervolino et al., 2009). Finally, 42% of the variance in hoarding symptoms was explained by genes in Taylor et al. (2010)'study, with genetic influences not varying as a function of gender nor age.

Shared environmental factors ranged from 3% (Iervolino et al., 2009) to 32% (Ivanov et al., 2013). Only one investigation found these influences to be significant in the development of hoarding, uniquely in adolescent girls (Ivanov et al., 2013); in contrast, shared environment played a largely negligible role in boys (Ivanov et al., 2013) as well as adult men and women (Iervolino et al., 2009; Iervolino et al., 2011; Lopez-Sola et al., 2014; Mathews et al., 2014; Taylor et al., 2010) and their effects could be dropped from the models without loss in fit in five out of six studies.

Finally, non-shared environmental factors and measurement error explained between 48% (Iervolino et al., 2009) and 75% (Lopez-Sola et al., 2014) of the variance in hoarding symptoms. Specifically, these factors explained 64% (95% CI 55-75%) and 75% (95% CI 63-87%) of the variance in hoarding in adolescent boys and adult men, respectively (Ivanov et al., 2013; Lopez-Sola et al., 2014). In female twin samples, these influences were estimated at 65% (95% CI 58-73%) in an adolescent sample; 48% (95% CI 43-55%) (Iervolino et al., 2009), 51% (95% CI 43-58%) (Iervolino et al., 2011), and 62% (95% CI 53-71%) in adult females (Lopez-Sola et al., 2014). Similar estimates were provided in studies reporting no gender differences in genetic and environmental factors to hoarding (Taylor et al., 2010; Mathews et al., 2014), where unique environmental factors and measurement error were estimated at 58% (95% CI 40-76%) and 66% (95% CI 56-77%) for adult male and female twins.

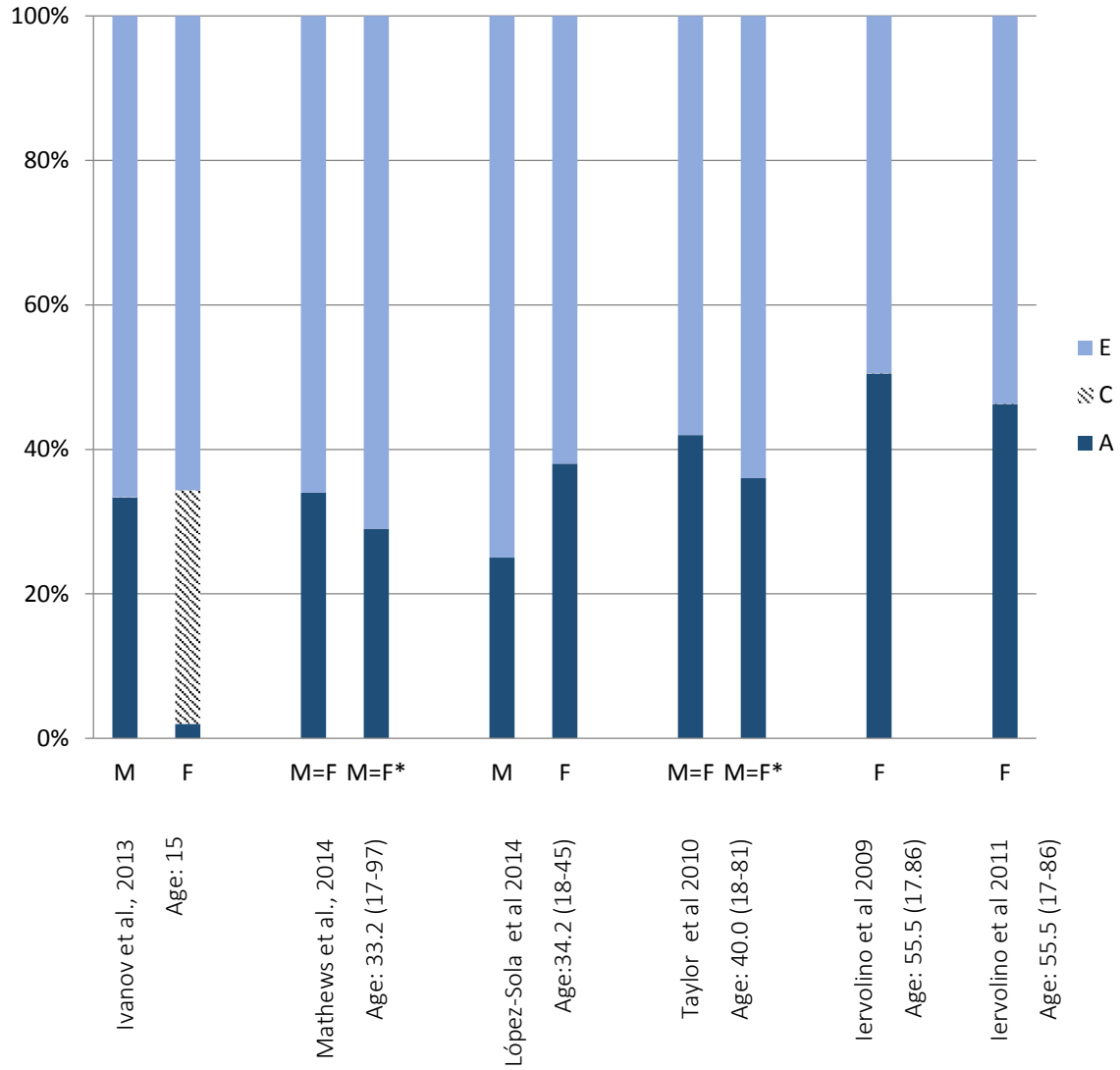
As shown in Table 3, Taylor et al (2010) and Mathews et al (2014) also replicated findings using more stringent cut-offs on the hearing measures to select participants with more severe symptoms, with similar results in variance components (Mathews et al., 2014; Taylor et al., 2010).

Table 3. Phenotypic variance explained by genetic (A), shared environmental (C), and unique environmental factors plus measurement error (E) for each study included, arranged by samples' mean age

	Best fitting model	Sex Differences	Male			Female			Male = Female		
			A	C	E	A	C	E	A	C	E
Ivanov et al., 2013	ACE	Yes (Quantitative)	.32 (.13-.44)	.04 (.0-.17)*	.64 (.55-.75)	.02 (.0-24)	.32 (.14-41)	.65 (.58-.73)	n.a.	n.a.	n.a.
Mathews et al., 2014	AE	N	-	-	-	-	-	-	.34 (.15-53) .29 (.17-.41) [B]	-	.66 (.56-.77) .71 (.65-.77) [B]
Lopez-Sola et al., 2014	AE	Yes (Quantitative)	.25 (.13-.37)	-	.75 (.63-.87)	.38 (.29-.47)	-	.62 (.53-.71)	n.a.	n.a.	n.a.
Taylor et al., 2010	AE	N	-	-	-	-	-	-	.42 (.24-.61) .36[A]	- -	.58 (.40-.76) .64[A]
Iervolino et al., 2009	AE	n.a.	n.a.	n.a.	n.a.	.49 (.30-57)	.03 (.0-.20)*	.48 (.43-.58)	n.a.	n.a.	n.a.
Iervolino et al., 2011	AE	n.a.	n.a.	n.a.	n.a.	.44 (.29-54)	.05 (.0-.17)*	.51 (.43-.58)	n.a.	n.a.	n.a.

*Abbreviations: n.a., not applicable; A, additive genetics; C, shared environmental factors; E, unique environmental factors plus measurement error; [A] Estimates calculated using more stringent cut-offs on the Obsessive-Compulsive Inventory. [B] Estimates calculated using more stringent cut-offs on the Hoarding Rating Scale Self-Report; *non-significant (p>.05).*

Figure 3. Phenotypic variance explained by genetic (A), shared environmental (C), and unique environmental factors plus measurement error (E) for each study included, arranged by samples' mean age



* *Estimates calculated using more stringent cut-offs on the hoarding measure*

5. DISCUSSION

Hoarding heritability estimates vary across twin studies and the reasons for this remain unclear. Recent twin studies have reported that genetic and environmental factors influencing hoarding symptoms may differ for males and females and across age groups, which in turn may be accounting for variation across studies (Ivanov et al., 2013; Lopez-Sola et al., 2014). We herein aimed to review and summarise for the first time the results of twin studies on hoarding, in view of examining the role of genetics versus environmental risk factors for hoarding and also shedding light on sex- and age-related differential effects in the familial transmission of hoarding symptoms. Results confirm hoarding symptoms as moderately heritable, with environmental factors playing an important role predisposing individuals to these symptoms. Albeit far from conclusive, we found some indication of differences in heritability of compulsive hoarding between men and women; particularly, the review highlighted age-related changes in hoarding heritability for females. However, more twin research is required to draw any firm conclusions on sex and age differences in the variance components to hoarding symptoms.

The quality of included studies was moderate to high for selection and methodological bias but overall poor for confounding bias. The methodological quality assessment of included studies highlighted the low percentage of descriptions on how missing data and confounding variables were assessed and controlled for, underling a risk for confounding bias across the available literature. This in turn would affect the heritability estimates; indeed, selection and confounding biases can influence MZ and DZ correlation estimates, resulting in an over- or under-estimation of heritability (Martin & Wilson, 1982). Given some evidence of gender and age effects in twin studies of hoardingthe quality assessment results emphasise the need to consider the statistical effects of age/sex and other confounding variables in any future examinations of this kind.

Collectively, the twin studies included in this review indicate that genetic factors play an important role in the aetiology of hoarding symptoms. Hoarding was found to be moderately heritable in adult men (25-42%) and women (range 34-49%); whilst also heritable in adolescence, the magnitude of genetic factors appeared to be significantly stronger in boys (32%) than in girls (2%). Although results on heritability estimates do not guarantee the success of subsequent gene mapping, they support the exploration of such approaches in future work. The search for specific genes in HD is in its infancy, most likely as a reflection of HD being recognized as a disorder separate from OCD only in recent years (Mataix-Cols et al., 2010). Nevertheless, a few molecular

genetic studies have been conducted, implicating regions on chromosomes 4, 5, 6, 14, 17, and 19 and a few genes previously implicated in obsessive–compulsive disorder (OCD) including *COMT, NTRK3*, and *SLC1A1* (Samuels et al., 2007a, b). Given the limited research and small sample sizes, firm conclusions on susceptibility genes for HD cannot yet be drawn at this stage; nonetheless, the review supports such research endeavours.

There seems to be some evidence of genetic influences accounting for a different amount of variance in hoarding for adolescence and adulthood, males and females. Firstly, hoarding heritability was lowest in adolescence girls and highest for female adult twins, suggesting an increase in genetic risk factors with age. This pattern was not observed for men, where heritability estimates were relatively similar across studies for adolescent and adult men. This observation could potentially account for the higher rates of hoarding reported in the literature of female relatives of hoarders and the increase in hoarding severity with age (Steketee et al., 2015). Second, albeit not consistently, sex differences were reported in some studies. Whilst two studies of twins aged and 17-97 and 17-81 found no evidence for sex-specific effects (Mathews et al., 2014; Taylor et al., 2010), two other investigations of younger twins aged 15 and 18-45 reported some differences in the *magnitude* of genetic influences on hoarding for males and females. The inconsistent findings highlight the possibility that gender differences may be more pronounced in younger individuals, and that sampling twins of a wide range of ages could hinder heritability differences across sexes. As discussed later on, given the limited number of studies so far, to examine developmentally dynamic sex-specific heritability changes, more twin studies are needed to shed further light on this issue. Taken together, however, the current review supports the need for genetic research for the identification of genes conferring risk to hoarding symptoms. The results furthermore highlight the potential benefit of taking sex- and age-specific effects into account in the search for predisposing genes to HD. As such, collecting large population-based samples and international collaboration will be paramount.

As previously shown, twin studies on hoarding also emphasise the importance of environmental factors in predisposing individuals to hoarding. Shared environmental influences played a negligible role on hoarding in all studies on adult twins (men and women) and one study on adolescent boys (Iervolino et al., 2009; Taylor et al., 2010; Iervolino et al., 2011; Ivanov et al., 2013; Lopez-Sola et al., 2014; Mathews et al., 2014). A striking different pattern however was observed in adolescent girls (Ivanov et al., 2013): at 15 years of age, shared environmental factors

showed an effect on hoarding symptoms, which disappeared or was not observed in adult female samples. Increases in heritability and decreases in shared environmental factors with age has been reported for other conditions, including OCD (Hur et al., 2008; Moore et al., 2010; Cornes et al., 2007; Mustanski et al., 2004) and has often been explained as an adult's greater control over their environment compared to adolescents. The fact that these influences were detected only for girls indicates that this teenage girls may be more vulnerable to familial environmental effects when compared to their male counterparts. Whilst more twin studies on younger samples are required to draw any firm conclusions, on the whole, the results suggest that familial environmental influences may be more important in determining hoarding behaviour among young females than genetic effects; it may be that these environmental circumstances are either more prevalent among girls and/or girls are more vulnerable to their effects. For males and older females the opposite trend applies, whereby genetic factors are more important than common environmental factors in predisposing to hoarding symptoms. Alternative explanations could explain the observed patterns of results however. Indeed, to date, only one twin study has been carried out in adolescence (Ivanov et al., 2013), which limits our ability to draw any firm conclusion; the study furthermore employed the HRS-SR, which was developed to reflect DSM-5 diagnostic criteria in adults. Very little is known about the clinical presentation of hoarding in young people, whether the use of the HRS-SR is a validated approach to reliably detect clinically significant hoarding symptoms in young people, and more generally, whether DSM-5 hoarding criteria apply in this age group. Collectively, results need to be interpreted cautiously and more research is needed to provide additional data to elucidate on age- and gender- effects in hoarding symptoms.

With regards to unique environmental factors on the other hand, all twin studies report approximately half or more of the variance in hoarding symptoms being accounted for by non-shared environmental factors, which includes measurement error. The 95% CI for unique environmental factors largely overlapped across studies, suggesting no significant variations with age and gender for unique environmental influences on hoarding.

Unfortunately, data on environmental risk factors for HD is fairly limited. Researchers have identified a number of factors that may contribute to the risk for developing HD; specifically, traumatic and stressful life events and trauma-related loss have been associated with the onset and severity of HD (Cromer et al., 2007; Landau et al., 2011; Grisham et al., 2006; Hartl et al., 2005; Timpano et al., 2011; Ayers et al., 2010; Tolin et al 2010). Whilst material deprivation has

also been implicated as an important risk factor for HD, recent investigations have been unable to confirm this association (Landau et al., 2011; Tolin et al., 2010). Overall, findings remain preliminary and require replication in much larger, epidemiological and longitudinal prospective studies. Taken together, however, these results support the crucial role of environmental factors and further examination and identification of these risk factors. Specifically, the results support the investigation of shared as well as unique environmental risk factors that confer risk to HD and highly encourage researchers to consider gender in the examination of such risk factors for hoarding. Identification of environmental risk factors will have important implications for designing intervention and prevention strategies, as these factors are potentially amenable to modification at this stage in comparison to genetic factors.

6. LIMITATIONS OF THE AVAILABLE RESEARCH AND FUTURE DIRECTIONS

To date, a total of four twin studies on hoarding included male and female twins, only one of which was carried out in adolescence. On the whole, twin studies on hoarding included samples that consisted predominantly of female Caucasian twins (78%), pooling hoarding data from twins across a wide range of ages (17 to 97). The review highlights the potential disadvantage of fitting single heritability statistics to data from twins from a wide range of ages given the suspected impact of age. Furthermore, all studies identified employed a cross-sectional design, estimating the magnitude of genetic and environmental influence of hoarding at a single time point. Ultimately, in order to fully assess the dynamic nature of heritability of hoarding and explore differences across the lifespan and gender, twin studies employing a longitudinal design are required.

Half of the included studies reported results from full ACE models; the remaining studies only reported best fitting AE models estimates. AE models overestimate heritability if a small contribution of shared environmental factors existed. Indeed, relatively large samples are needed (approximately 500 twins) to detect small shared environmental effects. Thus especially in smaller studies (e.g. Taylor et al., 2010), caution is needed when interpreting non-significant shared environmental factors as evidence that these factors are uninfluential to the development of hoarding. As most twin studies on hoarding include large samples, findings of non-significant shared environmental influences on hoarding is unlikely to be a statistical or power issue and more likely to reflect the negligible role of such factors in hoarding. Nevertheless, future twin

studies should consider the importance of reporting full ACE models as opposed to nested models. Equally important would be the standard use of sex-limitation models to assess gender differences in the heritability of hoarding, as this would ensure consistency and comparisons across studies.

Whilst this review summarises findings on genetic and environmental risk factors conferring risk for compulsive hoarding, considering gene-environmental interplays - which as with many other conditions are very likely to be involved in the emergence of hoarding – will be crucial in order to gather an in-depth understanding of this issue. The interaction between genes and environment has not been investigated yet; incorporating measures of environmental factors in future twin studies could be one avenue to explore gene-environment interactions. The implications of an interplay between genes and environment in the development of hoarding may imply that the action of genes could potentially be modified by changes in the environment and may therefore be of critical importance for prevention and intervention strategies for individuals with a familial predisposition to HD.

None of the studies were able to estimate how much of the unique environmental variance was due to measurement error as data on hoarding was measured at a single point in time. Future twin studies should use multiple assessments time points in order to clarify the extent to which 'pure' unique environmental factors contribute to hoarding symptoms and adjust heritability and unique environmental estimates accordingly. In addition to twin studies, results encourage future research employing different methodologies to prioritise the investigation of environmental risk factors predisposing individuals to HD, recruiting samples large enough to consider and detect gender and age differences. Future longitudinal studies as well as the study of MZ twins discordant for hoarding could be a fruitful avenue for examining the causative role of suspected environmental factors on hoarding, such as stressful life events, familial factors, and material deprivation.

Finally, the twin studies largely consisted of samples recruiting Caucasian populations from Europe, Australia, and Canada; it remains unclear how heritability and results on genetic influences on hoarding apply to other ethnic groups. More research would be useful in view of gathering a better understanding of genetic versus environmental risk factors for hoarding across different populations and ethnic backgrounds.

Another consideration pertains to the operationalisation of hoarding and the use of self-report measures to assess hoarding behaviours. The OCI-R and HRS-SR have been used across the 6 identified studies. The consistent use of two validated measures of hoarding symptoms, with evidence of their strong psychometric properties, is a positive finding. One could argue however that the use of two different measures may result in the measurement of slightly different behaviours/phenotypes, an increase in measurement error, and potentially contribute to the differences in estimates across studies. Although this remains a possibility, it is reassuring that genetic and environmental estimates for hoarding were markedly similar in a study administering the two different scales on the same subjects at different time points (Iervolino et al., 2009; 2011), suggesting HRS-RS and OCI-R broadly assess the same phenotype of interest.

On a related note, whilst the use of self-report measures as opposed to clinician-administered diagnostic interviews is a valid and widely employed method in behavioural genetics (Neale and Cardon 1992), it is limited by its inherent inability to determine diagnoses with absolute certainty. This raises three issues/limitations in itself: the use of self-report measures to assess hoarding could be problematic given evidence of hoarders' poor insight of their hoarding symptoms. Furthermore, using self-report measures, studies are unable to confidently determine whether hoarding symptoms were attributable to other mental or medical conditions (e.g. hoarding to OCD). Finally, the use of self-reported measures in adolescence have not been validated in adolescent populations. The limitation related to the use of self-reported measures therefore is important as it likely to increase measurement error and affect heritability estimate. Ultimately, therefore, future studies using psychiatric diagnostic interviews are warranted.

7. STRENGTHS AND LIMITATIONS

The current work should be interpreted in light of a number of strengths and limitations as below described.

This is the first systematic review of twin studies on hoarding to date, providing an overview of the results on aetiological risk factors for hoarding behaviour. The review was furthermore completed using a wide and systematic search of the literature, including the grey literature, in an attempt to avoid omitting any relevant data.

The objective of the current review was to investigate heritability of hoarding symptoms and attempt to shed light on any gender- and age-related effects on risk factors for hoarding. A limitation of the study is that despite the use a wide literature search approach, only six studies met inclusion criteria, two of which were carried out on the same twin sample albeit using different measures (Iervolino et al., 2009; Iervolino et al., 2011). While general conclusions on the heritability and role of the environment in predisposing individuals to hoarding could be drawn, the limited number of studies including male and female twins did not allow us to draw firm conclusions on the impact of age and gender on heritability estimates for hoarding at this stage. Further research is needed to provide additional data and clarify this issue further. Although beyond the scope of the current review, undertaking a meta-analysis might allow the estimation of genetic and environmental factors being more rigorously assessed; this in turn would allow a better examination and quantification of the reasons behind the variation in results in twin studies of hoarding.

Another limitation of the current study relates to the choice of the quality assessment tool. As recognized in a recent systematic review of quality assessment tools, to date, no 'gold standard' or general tool exists to assess risk of bias of behavioural genetic research. The choice of our tool therefore was largely dictated by what previous systematic reviews and meta-analyses of twin studies had used (Zeng et al., 2014); the scale was modified according to previous systematic reviews on twin studies (e.g. Zeng et al., 2014), with scant data on its validity and reliability for assessing the methodological quality of twin studies. The revised checklist furthermore only included one item to assess the risk of potential confounds, in relation to how confounding variables were measured and adjusted statistically for their impact on the outcomes. As such, the assessment of confounding risk could not be thoroughly assessed and it cannot be excluded that

the observed genetic/environmental estimates may therefore be the result of confounding variables. Notably, however, the assessment tool focuses on examining what are considered the three major bias components (ie selection, measurement, and confounding biases) and outweighs the benefits of developing a new assessment tool specifically for this study design. The lack of psychometric properties nevertheless remains one of the main limitation of the present systematic review.

Attempts to contact authors for additional or missing information were made (e.g. 95% CI), though not always successfully. The high risk of confounding bias observed in the included studies may be a reflection of the quality of the reporting as opposed to the quality of the study methods; this possibility cannot be excluded given the lack of contact with some of the authors.

Finally, the current study aimed to review and summarise findings from twin studies on compulsive hoarding. Findings will need to be interpreted in view of the general limitations of the twin design. One of the main assumptions of the twin method is that the extent of environmental influences shared between twin pairs does not differ as a function of zygosity, termed the 'Equal Environments Assumption' (EEA). Violations of this assumption (e.g., if twin pairs are treated more alike or exposed to more similar environments based on zygosity) can impact parameter estimates. Specifically, MZ correlations would be increased if MZ twins experience more similar environments than DZ twin pairs, leading to an overestimation of genetic influences. The effect of the EEA may work in the opposite direction, where shared environmental similarity is greater for DZ versus MZ pairs (e.g. due to the systematic separation of MZ twins into different classrooms, compared to DZ pairs), which would lead to increased DZ correlations, contributing to an overestimation of shared environmental influences (Rijsdijk & Sham, 2002). Another limitation related to the twin method is whether twins can be considered as representative of the general population, and as such can findings from twin samples generalise to singletons. There is evidence to suggest that compared to singletons, twin pairs tend to have lower birth weight, are more frequently associated with obstetric and pregnancy complications and born more prematurely (Rijsdijk & Sham, 2002). Moreover, twins tend to show delays in language attainment and cognitive ability, although this group difference is absent by middle childhood (Plomin et al., 2008). Finally, assortative mating arises when mate selection is not entirely random and has been cited as another major limitation of the twin method. Indeed, the effect of positive assortative mating contributes to greater DZ twin pair correlations (as the average genetic similarity of DZ

twins is increased, whereas MZ genetic similarity is already 100%), leading to reduced heritability estimates and increased shared environmental factors..

8. CONCLUSIONS

In conclusion, in the present review we aggregated the results of a number of previous twin studies on hoarding. In response to our primary objective, the review provides evidence for hoarding being moderately heritable, with environmental factors playing a significant role in predisposing individuals to these behaviours. Albeit far from conclusive, the pattern of results is indicative of gender- and age- related effects on the genetic transmission of hoarding symptoms: From the studies here included, there is some evidence of an increase in heritability from adolescence to early and late adulthood in women; genetic influence on male hoarding on the other hand appear overall stable across development. Shared environmental factors seem more important than genes in predisposing young females to develop hoarding behaviours, with genes potentially playing a more significant role later in age. Additional research however is necessary to provide any conclusive evidence; paramount are longitudinal twin studies to clarify developmental gender-specific risk factors for hoarding. Heritability calculation should pay particular attention to statistical methods which estimate genetic contribution, ensuring to report estimates for all three parameters (i.e. A, C, and E) rather than the AE model; this will help avoid overestimation of heritability in the presence of a small contribution of common shared factors and low statistical power. Indication of gender-specific risk factors for hoarding emphasises the need to examine the nature of these differences more closely in future research, and how they may vary by age.

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10. APPENDIX

Appendix 1. DSM-5 Diagnostic Criteria for Hoarding Disorder	
A.	Persistent difficulty discarting or parting with possessions, regarding of their actual value
B.	This difficulty is due to a perceived need to save the items and distress associated with discarding them
C.	The symptoms result in the accummulation of possessions that congest and clutter active living areas and substantially compromise their intended use. If living areas are uncluttered, it is only because of the intervention of third parties (e.g. family members, cleaners, authorities)
D.	The hoarding causes clinically significant distress or impairment in social, occupation, or other important areas of functioning (including maintaining a safe environmnetal for self and others)
E.	The hoarding is not attributable to another medical condition (e.g. brain injury, cerebrovascular disease, Prader-Willi syndrome)
F.	The hoarding is not better accounted for by the symptoms of another DSM-5 disorder (e.g hoarding due to obsessions in obsessive-compulsive disorder, decreased energy in major depressive disorder, delusions in schizophrenia or another psychotic disorder, cognitive deficits in dementia, restricted interests in autism spectrum disorder)

Appendix 2. Brief description of the disorders included in the new "*Obsessive-Compulsive and Related Disorders* (OCRDs)" chapter in DSM-5	
Obsessive-Compulsive Disorder (OCD)	OCD is characterised by intrusive and recurrent obsessional thoughts and by repetitive behaviours, leading to significant distress and/or functional impairment.
Body Dysmorphic Disorder (BDD)	BDD is marked by an xcessive preoccupation with a perceived defect in physical appearance, that is not observable or appears slight to others, and by repetitive behaviours in response to these concerns, leading to clinically significant distress and/or functional impairment.
Hoarding Disorder (HD)	HD refers to a persistent difficulty discarding or parting with possessions, due to a perceived need to save items and the distress associated with discarding them, resulting in clutter and causing clinically significant distress and/or impairment.
Thrichotillomania (TTM) (Hair-Pulling Disorder)	TTM is a mental condition whereby the person experiences a compulsive urge to pull one's own hair leading to noticeable hair loss, distress, and social and/or functional impairment.
Excoriation (Skin Picking) Disorder (SPD)	SPD is characterised by recurrent and repetitive picking of the skin resulting in noticeable tissue damage, and significant distress and/or impairment resulting from the picking.

Appendix 3. Database search string

Hoarding Disorder OR Hoarding OR Obsessive Hoarding OR Compulsive Hoarding OR Hoard*

AND

Genetics OR Genetic* OR Gene* OR Twin OR Twins OR Twin Study OR Heritability OR Inherence OR Family Study OR Family

Appendix 4. Data Extraction Form	
Author	
Year	
Country	
Source	
Sample size	
Mean age (range)	
Gender (% Female)	
Zygosity determinant	
Instrument	
Best fititng model	
Gender differences (type)	
Twin correlations	
MZ_{FEMALE} DZ_{FEMALE} MZ_{MALE} DZ_{MALE} $DZ_{OPPOSITE\ SEX}$	
% Variance	
A_{FEMALE} C_{FEMALE} E_{FEMALE} A_{MALE} C_{MALE} E_{MALE}	

Appendix 5. Quality assessment results for each included study						
Criteria	*Iervolino et al., 2009*	*Taylor et al., 2010*	*Iervolino et al., 2011*	*Ivanov et al., 2013*	*López-Sola et al., 2014*	*Mathews et al., 2014*
1. Define the source of information (survey, record review)	Y	Y	Y	Y	Y	Y
2. List inclusion and exclusion criteria for exposed and unexposed subjects (cases and controls) or refer to previous publications	Y	Y	Y	Y	Y	Y
3. Indicate time period used for identifying patients	N	N	N	Y	Y	Y
4. Describe any assessments undertaken for quality assurance purposes (e.g., test/retest of primary outcome measurements)	Y	Y	Y	Y	U	Y
5. Explain any patient exclusions from analysis	Y	U	Y	Y	Y	Y
6. Describe how confounding was assessed and/or controlled	U	N	N	U	N	N
7. If applicable, explain how missing data were handled in the analysis	N	U	N	N	N	N
8. Summarize patient response rates and completeness of data collection	Y	U	Y	Y	Y	Y

Abbreviations: Y, Yes (criteria met); N, No (criteria was not met); U, Unclear (authors did not specify)

Maternal and paternal accommodation of paediatric obsessive-compulsive disorder symptoms and its impact on treatment outcomes

MAIN RESEARCH PROJECT

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1. ABSTRACT

Background: Family accommodation (FA) refers to how parents modify their lives and behaviour to facilitate their child's obsessive-compulsive disorder (OCD) symptoms. The Family Accommodation Scale-Parent Report (FAS-PR) is the most commonly used assessment tool of FA in the field. Despite its common use, however, there has been no consistency in its scoring across studies. Furthermore, studies on FA have predominantly focused on maternal accommodation and no empirical studies have been carried out to consider how fathers respond or accommodate their child's OCD symptoms in comparisons to mothers.

Objectives: The current study assessed the factor structure of the FAS-PR. The study then aimed to compare the extent of FA between mothers and fathers. Finally, the study aimed to identify predictors of FA separately for mothers and fathers, and their differential association with child response to Cognitive Behaviour Therapy (CBT).

Methods: Mothers and fathers of children with OCD (N=209) were asked to independently complete the FAS-PR. Confirmatory factor analyses of alternative models used in the OCD literature were tested to examine the best fitting structure and scoring for the FAS-PR. T-test and chi-square analyses were used to compare the extent of FA of OCD symptoms between mothers and fathers. Using structural equation modelling (SEM), predictors of maternal and paternal FA and their impact on treatment outcomes were examined via regression models.

Results: A 12-items bi-factor FAS-PR structure fitted the data best. Mothers reported significantly higher levels of daily accommodation than fathers, with both reporting provision of reassurance, participation in rituals, and facilitation of avoidance as the most frequent types of accommodation. Though some differences were observed, predictors of both maternal and paternal accommodation were child's OCD symptom severity, emotional and behavioural difficulties, and parent psychopathology. As previously found, both maternal and paternal accommodation predicted post-treatment OCD severity. However, only father's involvement in OCD rituals predicted a significant treatment response (defined as a 35% reduction in OCD severity post-treatment), independently of whether mother were also engaging in FA or not.

Conclusions: The field would benefit from the use of a 12-items, 2 subscales FAS-PR to gain more meaningful and consistent insight into FA in OCD. Both mothers and fathers accommodate child OCD symptoms with high frequency, and in very similar ways. Although mothers accommodate

to a greater extent than fathers, fathers' involvement in ritual is a significant predictor of the child's treatment response. Results emphasise the need to consider the whole family system, including fathers, in understanding and treating childhood OCD. Clinical implications, future research directions, and the study's limitations are discussed.

2. INTRODUCTION

Obsessive-compulsive disorder (OCD) in children and young people is common, with prevalence estimates ranging between 1% - 4% in epidemiological studies (Flament et al., 1988, Heyman et al., 2001, Valleni-Basile et al., 1995, Zohar, 1999). The disorder is associated with marked impairment in functioning for the young person as well as significant caregiver burden and distress (Amir et al., 2000, Cooper et al., 1996, Calvocoressi et al., 1995). The term family accommodation (FA) has been specifically used in the OCD literature to refer to the involvement and participation of family members in an individual's OCD rituals (e.g. providing reassurance, providing items, assisting in avoidance, modifying routines or schedules). Indeed, young people with OCD are frequently unable to independently complete activities of daily living, or engage in activities shared by the family, and as a result the entire family routine can become disrupted. Consequently, relatives often become involved in a young person's rituals in an effort to reduce the impact of symptoms on child and family functioning.

2.1. Measurement of family accommodation in OCD

The Family Accommodation Scale- Parent Report (FAS-PR) (Cavalcoressi et al., 1995) has been commonly used by researchers in the field to assess the extent of FA in OCD (*Appendix 1*). Despite its widespread use, however, there still no agreement on how the FA-PR should be scored; to date, different methods have been employed (*Figure 1*). For example, some authors, whilst recognizing the existence of the *Distress* and *Consequence* subscales, opted for computing an overall accommodation (i.e. *Involvement*) score based on the first nine items of the *Participation* and *Modification* subscales (Peris et al 2008; Leibowitz et al 2014), mirroring the approach in the original version of the FAS (Calvocoressi et al 1995). A second approach has been to use all the 13 items to create a total FAS-PR *Total* score (Boeding et al., 2013; Merlo et al., 2013; Peris et al., 2008; Storch et al., 2007; Torres et al., 2012). A similar third approach has been to create an overall accommodation score by lumping together only the first nine items, which focus specifically on parental involvement in the child's OCD rituals (Caporino et al 2012; Boeding et al 2013). Finally, Flessner and colleagues (2009) performed the first – and sole existing to date – exploratory factor analysis of the FAS-PR in a sample of 96 youth with OCD. They found a two-factor solution involving a first factor of *Involvement in Compulsions* (items 1-3 and 11-13) and a second factor of *Avoidance of triggers* (items 4-9), with both factors forming a *Total* score; the item pertaining to parental distress (item 10) was removed. This scoring version has been tested specifically in an adolescent OCD sample and has been adopted by others in more recent

investigations of FA in OCD (Bipeta et al., 2013; Flessner et al., 2011). Although factor analyses and rationale approaches have come up with different structures in both adult and youth samples, to date, no studies have formally compared these models with each other. Given the impact of FA, it is important to examine the scale's factor structure in view of supporting an adequate assessment of FA in OCD.

2.2. Clinical importance of family accommodation of OCD symptoms

To-date, a handful number of studies examined FA in childhood OCD largely using the FAS-PR and its various scoring methods (Bipeta et al., 2013, Caporino et al., 2012, Flessner et al., 2011, Futh et al., 2012, Garcia et al., 2010, Lebowitz et al., 2014, Merlo et al., 2009, Peris et al., 2008, Storch et al., 2007). Most of these have reported high rates of accommodation among families of pediatric OCD sufferers, with results indicating that between 60-96% of relatives assist or modify their behaviour to accommodate their child's OCD symptoms (Bipeta et al., 2013, Caporino et al., 2012, Flessner et al., 2011, Futh et al., 2012, Garcia et al., 2010, Lebowitz et al., 2014, Merlo et al., 2009, Peris et al., 2008, Shafran et al., 1995, Stewart et al., 2008, Storch et al., 2007). In addition to being common, various child- and parent-level factors have been found to predict parental accommodation, including OCD symptom severity, functional impairment, child's internalising and externalising symptoms, and parent psychopathology (i.e. maternal anxiety and depression) (Caporino et al., 2012, Flessner et al., 2011, Futh et al., 2012, Lebowitz et al., 2014, Merlo et al., 2009, Peris et al 2008, Stewart et al., 2008, Storch et al., 2007). A handful of studies across the pediatric and adult OCD literature have also observed an association between FA and treatment outcomes (Ferrao et al., 2006, Garcia et al., 2010, Merlo et al., 2009), albeit not all studies (e.g. Peris et al., 2008). Specifically, lower FA scores pre-treatment and greater reduction in FA post-treatment were associated with better outcomes as indicated by lower OCD severity scores at the end of treatment (Ferrao et al., 2006; Merlo et al., 2009; Garcia et al., 2010). Whilst the causal direction of these findings cannot be inferred, overall, the results lend some empirical support to the theoretical CBT framework, according to which FA hinders CBT effectiveness by reinforcing fear and avoidance (Peris et al., 2008; Storch et al., 2007). Whilst cognitive-behavioural therapy (CBT) is the recommended first-line treatment in youths with OCD (NICE, 2005), it achieves a success rate of approximately 60-70%, and patients (even treatment responders) are rarely asymptomatic after treatment. This means that new better treatments are needed to increase that success rate; with this in mind and given the above association, FA ought to receive greater attention as a potential target for improving OCD treatment outcomes.

2.3. Maternal versus paternal accommodation of OCD symptoms

Despite the increased interest in understanding factors that drive or promote FA, the extent to which patterns of accommodation vary between family members - and specifically between mothers and fathers of young people with OCD - remains unclear. Evidence suggests that mothers and fathers interact differently with their children, with fathers contributing uniquely to their child's behavioural and psychological development across various mental health diagnoses (Lewis & Lamb, 2003; Ramchandani et al., 2005; Ramchandani et al., 2013). The role or response of fathers to their child' OCD symptoms and treatment however is a largely neglected area of research. Indeed, the available OCD literature have either only involved one parent (most commonly the mother) or clustered together different kinship of family members within the same study. It is possible that there may be a differential response between family relatives to a child's OCD symptoms. For instance, in the adult OCD literature, it has been found that spouses/partners endorse significantly higher FA scores in comparisons to other family members, such as parents, siblings, children, and cousins, though no differences were noticed between mothers and fathers (Gomes et al., 2014). Another study has uniquely examined differences between mothers and fathers in response to their child's OCD symptoms (Futh et al., 2012). This qualitative study found no parental differences in understanding, narrative, coping, and distress associated with family accommodation, however - as the author themselves note - the self-reported diagnostic status of the children significantly limited the generalizability of their findings (Futh et al., 2012). To our knowledge, no study has empirically examined maternal and paternal accommodation of symptoms separately in a paediatric OCD sample. The question as to how FA differs between mothers and fathers and how these differences may impact or interact with the child' symptoms and treatment therefore remains to be addressed. Considering the paternal perspective and how fathers respond or accommodate their child's OCD symptoms can provide a greater understanding of FA in all its facets and help tailoring any clinical intervention accordingly.

3. AIMS OF THE CURRENT PROJECT

Given the impact of family processes in the presentation and treatment of paediatric OCD and the paucity of studies including fathers, the current study aimed to extend pre-existing work by examining maternal and paternal FA of OCD symptoms. Below is a summary of the main aims and hypotheses:

Aim 1: To examine the structure of FA – as measured with the Family Accommodation Scale Parent Report (FAS-PR) - by formally comparing the different models employed in the literature for the first time and see whether the same structure holds for mothers and fathers.
Hypothesis: A two-factor solution model (i.e. Avoidance of triggers and Involvement in compulsions) was hypothesised to fit our data best, both for mothers and fathers.

Aim 2: To assess whether the pattern and severity of FA of OCD symptoms differs between mothers and fathers.
Null hypothesis: no statistical differences are expected in the extent of FA between mothers and fathers.
Alternative hypothesis: statistically significant differences are expected in the extent of FA between mothers and fathers.

Aim 3: To compare predictors of FA of OCD symptoms separately for mothers and fathers.
Null hypothesis: OCD symptom severity and parent psychopathology would not predict FA.
Alternative hypothesis: OCD symptom severity and parent psychopathology predict parental accommodation.

Aim 4: To examine the association between FA, treatment outcomes.
Null hypothesis: maternal and paternal FA do not predict CBT treatment outcomes
Alternative hypothesis: maternal and paternal FA significantly predict CBT treatment outcomes

3.1. Power calculations

Power calculations were completed using GPower. Based on data from studies on FA in pediatric OCD (Flessner et al., 2009; Flessner 2011), a sample size calculation showed that in order to detect a clinically significant difference (i.e. 10%) between mothers and fathers on the primary

outcome measure (FAS-PR), at alpha 5% and 80% power, a sample of 81 parent pairs was required. A correlation between child- or parent-variables and FAS-PR scores of 0.3 (or above) has been observed in the study by Flessner et al., 2011 on predictors of FA in pediatric OCD; this is equivalent to a 9% explained variance. To account for 9% variance, at alpha 5% and 80% power, a sample of 82 mothers was estimated. The same sample size was assumed for investigation of predictors of paternal FA, given no prior investigations of FA has been completed on fathers. Finally, the power calculations to examine whether FA predicted treatment outcomes was based on a study looking at treatment outcomes and FA in pediatric OCD (Merlo et al., 2009); to obtain a significant partial r^2 of 0.17 as observed in this study, at alpha = 0.05 and 80% power, a sample of 55 mothers was calculated. The same sample size was assumed for paternal FA. Based on the above power calculations, the study aimed collect data from at least 100 parents of children and adolescents with OCD, to allow for drop-outs or incomplete data.

4. METHODS

4.1. Participants

The sample consisted of 209 children and adolescents aged 7- 18 years meeting diagnostic criteria for OCD and their parents. One hundred and twenty four participants (n=124; 59.3%) received CBT at the clinic and had post-treatment data available; the remaining patients were referred for assessment and treatment recommendations only or post-treatment data was missing (e.g. currently in treatment). There were no significant differences between participants who were treated at the clinic and those who were not with respect to age, gender, and OCD severity (all *p*>.05). Demographic and clinical characteristics of the sample are summarized in *Table 1*.

Table 1. Demographic and Clinical Characteristics of the total sample (N=209), including N=124 receiving treatment and N=85 attending for assessment only

Variable	N =209(%)	N =124 (%)	N =85 (%)
Gender			
Male	118 (56.5)	73 (58.9)	45 (52.9)
Female	91 (43.5)	51 (41.1)	40 (47.1)
Variable	**Mean (SD)**	**Mean (SD)**	**Mean (SD)**
Age at assessment	14.10 (2.38)	14.29 (2.18)	13.82 (2.64)
Age of OCD onset	10.42 (3.13)	10.72 (3.17)	9.96 (3.01)
CY-BOCS total score	27.00 (5.25)	26.78 (5.12)	27.32 (5.44)
Obsessions	13.14 (2.82)	13.01 (2.75)	13.33 (2.92)
Compulsions	13.86 (2.74)	13.77 (2.67)	13.99 (2.85)
CGAS score	45.68 (10.69)	47.45 (11.10)	42.57 (9.21)
BDI-Y Total score	20.83 (10.95)	20.00 (10.24)	22.14 (11.94)

Abbreviations: SD, standard deviations; OCD, Obsessive Compulsive Disorder; CY-BOCS, Children Yale-Brown Obsessive Compulsive Scale; CGAS, Children's Global Assessment Scale, Beck Depression Inventory for Youth.

4.2. Measures

Clinician-administered measures

Children's Yale-Brown Obsessive-Compulsive Scale (CY-BOCS) (Scahill et al., 1997)*:* the CY-BOCS is a clinician-rated semi-structured interview for assessment of paediatric OCD symptom severity, with sound psychometric properties (Scahill *et al.*, 1997; Storch et al., 2004). It includes an OCD symptom checklist followed by 10 items assessing OCD severity; severity scores range from 0 to 40. Scores of 16-23, 24-31, and 32 to 40 are indicative of moderate, severe, and extreme OCD severity, respectively (Scahill et al., 2007). In the current study, the CYBOCS demonstrated good internal consistency (Cronbach's α = .85).

Children's Global Assessment Scale (CGAS) (Shaffer et al., 1983)*:* the CGAS is a validated and reliable measure of severity of disturbance and adequacy of social functioning. The scale ranges from 1-100, with 1 representing the most impaired child and 100 representing the healthiest. Scores above 70 represent healthy functioning. The CGAS has shown reliability between raters and across time, and was used in the current study as a clinician-rated measure of functional impairment.

Parent- and Child- Self-report measures

Family Accommodation Scale –Parent Report (FAS-PR): the FAS-PR is a modified version of the semi-structured, clinician-administered FAS (Calvocoressi et al 1995). It is a parent-report 13-item measure that assesses the degree to which parents accommodate their child's OCD symptoms (*Appendix 1*). The FAS-PR measures both the behavioural involvement of family members in the child`s OCD (e.g., modification of daily routines, participation in rituals) and the level of family distress and disruption associated with this involvement. Individual items are rated on a 5-point Likert scale ranging from 0 (never) to 4 (daily). In the present study, both mothers and fathers were asked to complete the FAS-PR. Cronbach's alpha for the present sample were .91 for mothers and .92 for fathers.

Beck Depression Inventory for Youth (BDI-Y) (Beck et al., 1962; Beck et al., 2005)*:* The BDI-Y is a widely-used 21 item self-report measure for depressive symptoms, which has good internal consistency and test-criterion validity (Beck *et al.,* 2001). Total raw scores range from 0 to 63, with higher scores indicating greater symptom severity; scores above 14 are suggestive of moderate depressive symptoms and a cut-off of 29 or higher is indicative of severe depression (Erford & Muller, 2012). Cronbach's α for the BDI-Y in the present study was .92.

Depression Anxiety Stress Scale (DASS) (Lovibond et al., 1995)*:* The DASS is a 42-item self-report measure of parental negative emotional symptoms, with 14 items within each subscale assessing symptoms of Depression, Anxiety and Stress; scores on each subscale range from 0-42, with

higher scores indicating higher levels of distress. Parents rated the extent to which they have experienced the symptom over the past week on a four point severity/frequency scale. The measure provides a score for each of the three subscales in addition to a total score, indicative of general negative emotional distress. The measure has UK-based normative data and good psychometric properties. In the present study, the DASS was completed by both mothers and fathers; cronbach's α values were .97 for both mothers and fathers.

Strengths and Difficulties Questionnaire (SDQ) (Goodman, 1997; Goodman, 2001)*:* the SDQ is a 25-item questionnaire incorporating 5 subscales capturing emotional (items 1-5), conduct, (items 6-10), hyperactivity/inattention (items 11-15), peer problems (items 16-20), and pro-social behaviour (items 21-25). Total scores range from 0 to 40, with prosocial behaviour subscale scores excluded from total score calculations (Goodman, 2001). The measure is widely used across a range of clinical settings, and has been shown to have good psychometric properties, including good internal consistency and retest stability, with elevated scores predictive of psychiatric diagnosis (Goodman, 2001). Cronbach's α values was .62 for parent-reported SDQ.

4.3. Procedure

All patients and families attended an initial assessment of approximately three hours with a multidisciplinary specialist team. The majority of young people referred were seen for assessment only with the view of providing recommendations for treatment to treating clinicians in generic child mental health services. Given the location of the specialist service in London, only those who lived locally enough or who found travel to the clinic feasible were treated at the clinic. Accordingly, a total of 124 OCD patients (59.3%) received CBT treatment at the clinic (mean number of CBT sessions = 15.05, SD=5.5), delivered by trained therapists or by trainees under close supervision from experienced therapists. The CBT intervention was protocol-driven (see Nakatani et al, 2011) and consisted of weekly sessions incorporating psycho-education on OCD and anxiety, exposure and response prevention (E/RP), and relapse prevention. Approximately a third of those receiving CBT (35.9%) were also on SSRI medication; in most cases medication had reached a stable dose before CBT commenced. Those receiving medication were more likely to present with more severe OCD symptoms and more impaired scores on measures of social functioning (CGAS) and depression (BDI-Y) ($p<0.01$). Clinician- and self-report measures were administered at different time-points (at assessment or pre-treatment, post-treatment, and at follow-up). The study utilizes data that is routinely collected from families attending the N&S paediatric OCD Clinic. The clinic's research assistants were in charge of sending out and collecting

child- and parent-report measures as part of the clinic's standard practice. Research assistants, qualified as well as trainee clinical psychologists - including myself - completed the clinician-administered measures at regular time points. The current study received Clinical Audit Ethical approval (by CAMHS Audit Committee) on 8.10.2014 and followed relevant research governance as outlined in the ethics approval form.

4.4. Statistical Analyses

For our first aim, we used confirmatory factor analysis (CFA) to evaluate and compare the relative fit of four alternative factor structures for the FAS-PR used across the OCD literature in both mothers and fathers separately (*Figure 1*). CFA is a type of factor analysis that tests a pre-established structure between observed and latent factors. The objective of CFA is to test whether the data fit a hypothesized measurement model. All assumptions of the CFA in this study were met, including assumptions related to study design (i.e. sample size of at least 200 individuals, a priori model specification, and independence of observations), and those related the nature of the data, that is the assumption of multivariate normality, assessed via boxplot inspection and by the shapiro-wilk estimate value (.97 and .95 for mother and fathers FAS-PR, respectively) falling in the acceptable range for normality (±2.00). The first model (hereafter *Model 1*) was a bi-factorial model with a four-first order group factors or subscales (*Participation*, items 1-5; *Modification*, items 6-9; *Distress*, item 10; and *Consequences*, items 11-13) and a general factor (*Total* score) loading on *Participation* and *Modification* items (Lebowitz et al., 2014; Peris et al., 2008). The second model (*Model 2*) was a unidimensional model with a single factor lumping all 13 items (Boeding et al., 2013; Merlo et al., 2013; Peris et al., 2008; Storch et al., 2007; Torres et al., 2012). The third model (*Model 3*) was also a unidimensional model with a single factor lumping the first 9 items of the FAS-PR (Caporino et al 2012; Boeding et al 2013). The fourth and final model (hereafter *Model 4*) was a bi-factor model with two-first order group factors (*Avoidance of triggers,* items 4-9; *Involvement in compulsions*, items 1-3 and 11-13) and a general factor (Total score) loading on 12 items; the distress item (item 10) was not included (Flessner et al 2009; Flessner al., 2011; Bipeta et al 2013). The four models are depicted in *Figure 1*. We performed CFA in MPlus version 7 (Muthen and Muthen, 2012), using maximum-likelihood estimation with robust standard errors (MLR). We followed common practice in reporting multiple indices of model fit, namely the Comparative Fit Index (CFI), the Tucker Lewis Index (TLI), and the Root Mean Square Error of Approximation (RMSEA) (Hu & Bentler, 1999; Brown, 2006;

Hopoer et al 2008). To consider a model as showing 'acceptable' fit, a CFI>0.90, TLI>0.90, and RMSEA<0.08 is typically required; A CFI>0.95, TLI>0.95, and RMSEA<0.06 (Brown, 2006) is indicative of a model showing a 'good' fit. When two or more models show acceptable/good fit, the Akaike Information Criteria (AIC) and the Bayesian Information Criteria (BIC) provide an additional indication of model fitness, with lower values indicating better fit (Burnham & Anderson, 2004; Levy & Hancock, 2007). The best fitting FAS-PR model/structure was then employed for all subsequent analyses.

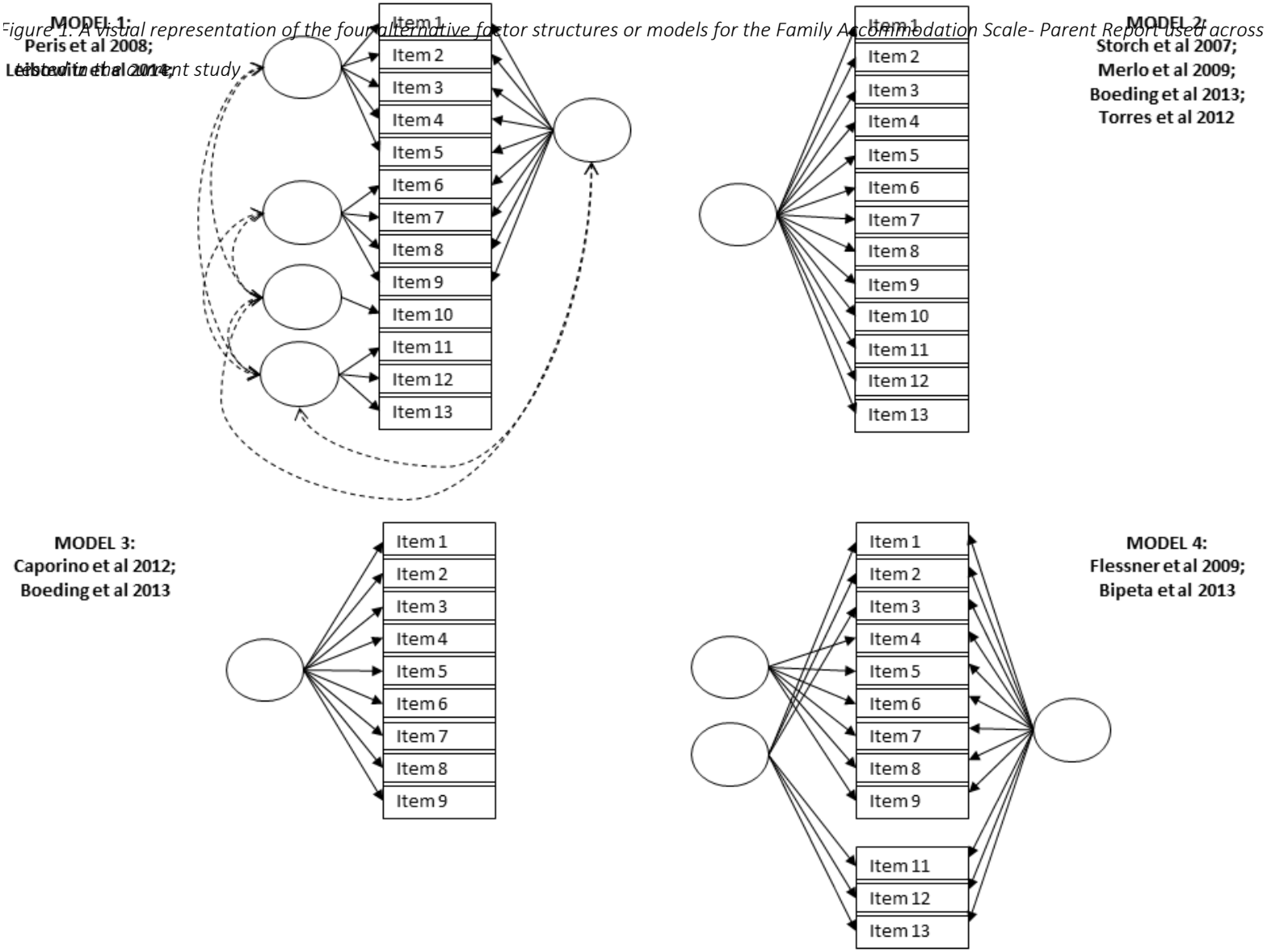
Our second aim was to compare the pattern and extent of maternal and paternal accommodation of OCD symptoms. To this end, comparisons in FAS scores between mothers and fathers were performed using Wilcoxon sign rank test for FAS-PR scores and chi-square tests for individual items. Wilcoxon sign rank test is a non-parametric test to compare two related samples. The data was assessed for violation of assumptions prior to analysis, including paired samples measured at the ordinal level, independence of observations (i.e. the observations were randomly drawn from the population), symmetrical distribution of differences; the first and second assumptions were met by design, whilst symmetrical distributions was confirmed by the inspection of the separate distributions being similar in shape and values (mother FAS median score=24.00; father FAS median score= 21.00; mother and father FAS interquartile range = 20 and 22 respectively).

Structural equation modeling (SEM) was used to address *Aims 3* and *4*. Specifically, the third aim of the study was to examine predictors of FA separately for mother and fathers in order to assess which demographic and clinical variables were differentially related to maternal versus paternal accommodation. Correlation analyses were first employed to select variables for inclusion in the regression models (i.e. predictors), with $p<.05$ as the criterion for entry; variables of interest included baseline OCD symptom severity (CY-BOCS score), child psychopathology (SDQ total score), depressive symptoms (BDI score), parent negative emotional symptoms (DASS score), overall general functioning (CGAS), as well as gender, age and duration of illness. Using SEM in MPlus, a series of linear regression analyses were then performed first with maternal and paternal FAS scores as outcome separately, and then including both as outcome in the same model.

The paper's final aim was to examine whether maternal and paternal FA predicted treatment outcomes and, if so, whether they had an independent effect on these outcomes. Using SEM and consistently with previous studies of this kind, this final aim was first tested with post-treatment CY-BOCS score entered as the dependent variable, maternal and paternal FAS scores as

predictors, and pre-treatment CY-BOCS as a covariate to control for initial symptom severity. Analyses were then repeated using 'treatment response', defined as a 35% reduction in CYBOCS scores from pre- to post-treatment (Mataix-Cols et al., 2016) as the dependent variable. Assumptions pertaining to linear regressions were tested prior to completing analyses pertaining to Aim 3 and 4 and met; indeed, the boxplot and scatterplots suggested a normal distribution shape of the errors, with no significant outliers observed. Taken together, inspection of scatterplots revealed that as values of the independent variables increased, those of dependent variables also increased, indicating that the assumption of linearity was reasonable. The Durbin-Watson (DW) statistic was used to evaluate independence of errors, with values falling in the acceptable range (>1.0) suggesting independence of observations (Field, 2009). Finally, the assumption of multicollinearity was also met, with data indicating correlations among independent variables of .7 or less.

Figure 1. A visual representation of the four alternative factor structures or models for the Family Accommodation Scale- Parent Report used across the OCD literature and current study.



5. RESULTS

5.2. Aim 1: To examine the structure of FAS

As shown in *Table 2*, in mothers CFI, TLI, and RMSEA showed good fit for *Model 1* and *Model 4,* and CFI was acceptable for the unidimensional *Model 3*. In the case of father-reported FAS-PR, CFI, TLI and RMSEA showed a good fit for *Model 1,* and for *Model 4*, CFI was good and TLI and RMSEA were acceptable. Overall, *Model* 1 and *Model* 4 showed the best fit to the data for both mothers and fathers. In contrast, unidimensional models (i.e. Models 2 and 3) of FAS-PR showed the poorest fit to the data. Based on the comparison of AIC and BIC between Models 1 and 4, *Model 4* was selected for having a lower AIC and BIC as well as for being more parsimonious model.

Table 2. Model fit in Confirmatory Factor Analyses of the different FAS models.

Source	Model	χ^2	df	*p*	CFI	TLI	RMSEA	AIC	BIC
Mother	*1*	66.451	54	0.119	0.990	0.986	0.033	8280.17	8447.29
	2	241.644	65	<.000	0.859	0.831	0.114	8462.10	8592.45
	3	97.721	27	<.000	0.906	0.874	0.112	5980.10	6070.34
	4	58.148	42	0.049	0.986	0.978	0.043	7713.91	7874.34
Father	*1*	91.872	54	0.001	0.970	0.957	0.058	8082.67	8249.79
	2	257.979	65	<.001	0.848	0.818	0.119	8278.97	8409.32
	3	106.840	27	<.001	0.889	0.852	0.119	5838.64	5928.89
	4	78.993	42	0.005	0.967	0.949	0.065	7559.92	7720.35

Abbreviations: χ2, chi-square; df, degrees of freedom; p= p value; CFI= Comparative Fit Index; TFL, Tucker Lewis Index; RMSEA, Root Mean Square Error of Approximation; AIC, Akaike Information Criteria; BIC, Bayesian Information Criteria.

10.1. 5.2. Aim 2: To examine the extent of maternal versus paternal accommodation of OCD symptoms

Parental accommodation was common; both mothers (98.1%) and fathers (97.6%) reported engaging in some form of accommodation of their child's OCD symptoms daily. In addition, there was a high correlation between mother and father FAS-PR total score (r=0.74, 95%CI 0.67-0.80,

p=<0.0001). As shown in *Table 3* and *Figure 2*, similar patterns of accommodation were observed for both types of parents, with provision of reassurance, participation in rituals, and facilitation of avoidance being endorsed as the most frequent types of accommodating behaviours by both parents. However, results also showed that mothers scored higher on all items. Relative to fathers, mothers reported significantly higher rates of accommodation of their child's OCD symptoms on the FAS-PR Total score [z = 7.071, p < 0.0001]. Mothers also reported higher scores in *Avoidance of triggers* [z = 5.083, p < 0.0001] and *Involvement in compulsions* [z =7.511, p < 0.0001] subscale. Percentages of items endorsed daily and mean levels of FAS items for mothers and fathers are reported in *Table 2* and *Figure 2*, respectively.

Table 3. Percentage of mothers and fathers endorsing each of the FAS-PR accommodation items daily.

FAS-PR items	Frequency of daily accommodation			
	Mother	Father	χ^2	*p*-value
1. Providing reassurance	*61.7%*	*36.4%*	*31.90*	*<.001*
2. Providing items for compulsive behaviours	26.3%	13.9%	36.90	*<.001*
3. Participating in behaviour related to compulsions	*44.5%*	*27.8%*	*39.39*	*<.001*
4. Assisting in avoidance	*42.6%*	*28.2%*	*27.51*	*<.001*
5. Modifying personal routine due to OCD	13.4%	11.0%	70.27	*<.001*
6. Modifying family routines due to OCD	17.7%	14.4%	57.64	*<.001*
7. Assuming responsibilities for child	11.0%	8.6%	5.66	0.017
8. Modifying work schedule due to OCD	16.7%	8.6%	10.84	0.001
9. Modifying leisure activities due to OCD	16.7%	11.0%	36.09	*<.001*
10. Own distress caused from accommodating	14.8	7.2%	4.38	0.036
11. Child distressed/anxious when not assisted	34.9%	24.4%	56.17	*<.001*
12. Child angry/abusive when not assisted	29.2%	21.1%	36.36	*<.001*
13. If unassisted, child spends increased time ritualising	20.6%	14.8%	36.93	*<.001*

Abbreviations: FAS-PR, Family Accommodation Scale Parent Report; χ^2, chi-square; Items in italics denote most frequently endorsed items; p <0.05.

Figure 2. Mean item scores on the Family Accommodation Scale for mothers (blue) and fathers (red)

10.2. 5.3. Aim 3: To examine and compare predictors of maternal and paternal accommodation of OCD symptoms

No significant correlations were found between mothers' or fathers' FAS scores and child demographic variables (age, gender, duration of illness); these variables were therefore excluded as possible predictors from the regression analyses. In contrast, OCD symptom severity (CY-BOCS), general functioning (CGAS), the child emotional and behavioural difficulties (SDQ), depressive symptoms (BDI-Y), and mothers' and fathers' DASS total scores were significantly correlated with mothers' and fathers' FAS; these variables were therefore retained as predictors and entered into a regression analyses.

The model predicting maternal FAS accounted for 40% of the variance (R^2 = 0.396), with OCD symptom severity (β = .28, p<0.001), mother DASS (β = .34, p< 0.001), father DASS (β = .15, p= 0.045.), and SDQ total score (β = .16, p= 0.028) making a significant contribution. With regards to paternal accommodation, the model accounted for 34% of the variance (R^2 = 0.336), with OCD symptom severity (β = .21, p=0.008), paternal DASS (β = .32, p< .001), and SDQ total score (β = .16, p= 0.045) making a significant contribution. BDI and CGAS did not predict maternal nor paternal accommodation of OCD symptoms.

When the association between mother and father FAS was taken into account, predictors explained 40% (R^2 = 0.397) of the variance of mother FAS and 33% (R^2 = 0.331) of the variance of father FAS. In this model, maternal and paternal accommodation were significantly predicted by the same clinical indicators as when predicted separately. That is, mother FAS was significantly predicted by OCD symptom severity (β = .28, p<0.001), mother DASS (β = .34, p< 0.001), father DASS (β = .15, p= 0.045), and SDQ total score (β = .16, p= 0.031). By contrast, father FAS was significantly predicted by OCD symptom severity (β = .21, p=0.010), paternal DASS (β = .33, p< .001), and SDQ total score (β = .16, p= 0.042). The strength of the predictions was the same as when maternal and parental FAS were predicted separately, suggesting that these clinical indicators independently predicted mothers' and fathers' symptom accommodation and were not influenced by the high correlation between the two informants.

Finally, additional analyses were carried out using the FAS-PR subscales. The same patterns of predictors were observed for maternal and paternal involvement in rituals and avoidance of

triggers: predictors explained 30% of the variance (R^2=.298) of maternal and 25% of paternal (R^2=.249) involvement in rituals, respectively; predictors explained 41% (R^2=.407) of variance of maternal and 38% (R^2=.378) of paternal avoidance of triggers, respectively.

10.3. 5.4. Aim 4: Parental accommodation and treatment outcome

A subset of treatment completers from the total sample were used (n = 124) to examine the impact of maternal and paternal on treatment outcome.

When using CYBOCS post-treatment scores, both maternal (β = -.26, *p*= .001) and paternal (β = .15, *p*= .005) accommodation significantly predicted post-treatment CYBOCS scores when controlling for pre-treatment OCD severity (i.e. baseline CYBOCS scores). However, only paternal FAS-PR (β = -.29, *p*= .026) and baseline OCD severity (β = -.19, *p*= .035) predicted 'treatment response', defined as a significant reduction of 35% or more in CYBOCS scores pre- to post-treatment (Mataix-Cols et al., 2015), while maternal accommodation did not (β = .02, *p*= .855). This model accounted for 14.2% of the variance (adjusted R^2=14.2). Analyses were repeated using the two FAS-PR subscales (i.e. *Avoidance of Triggers* and *Involvement in Compulsions*) to examine their predictive effect on treatment response; fathers' involvement in compulsion was a significant predictor of treatment response (β = -.43, *p*=.006) alongside baseline OCD severity (β = -.18-, *p*=.043); fathers' avoidance of triggers (β =.14, *p*=.381), maternal involvement in compulsions (β =.18, *p*=212)and avoidance scores (β -.16=, *p*=.305) on the other hand did not make a significant contribution.

6. DISCUSSION

The majority of studies on FA in OCD have relied on the use of the FAS-PR as the gold standard measure for the assessment of FA in OCD; currently, there is no agreement on how the FAS-PR should be scored and no studies have formally compared the different approaches used across the OCD literature. This study sought to examine the scale's factor structure, by formally comparing the different models employed in the literature for the first time. Moreover, to date, only two studies examined FA in relation to kindship of relatives (Gomes et al., 2014; Futh et al., 2012). Fathers' response to their child' OCD symptoms therefore remains a largely neglected area of research. Consequently, the present project examined patterns and predictors of parental accommodation of OCD symptoms in young people and its association with treatment, with a specific and unique emphasis on investigating and directly comparing maternal and paternal accommodation. The main findings of the current study will be discussed in the following paragraphs, followed by a discussion of the clinical implications, limitations, and future research directions.

To the best of our knowledge, this is the first study to test and compare the fit of different FAS-PR models used in the literature. Out of the four models used in the OCD literature and compared in the current study, the bi-factor 12-items FAS-PR model, incorporating two subscales (*Avoidance of Triggers* and *Involvement in Compulsions*) and a *Total* FAS-PR score, fitted the data best; as hypothesized, this model yielded the best fit for both mothers and fathers. This model was initially generated from the only exploratory factor analysis of the FAS-PR on a sample of youths with OCD (Flessner et al., 2009) and used in subsequent studies thereafter (Flessner et al., 2011; Bipeta et al., 2013). Taken together, our findings support Flessner et al (2009)'s FAS-PR model as having the strongest factor analytic support to date. Although researchers often create a FAS-PR total score by summing the thirteen or first nine FAS-PR items (*Model 2* and *3*) when assessing FA in OCD (Boeding et al., 2013; Caporino et al., 2012; Merlo et al., 2013; Peris et al., 2008; Storch et al., 2007; Torres et al., 2012), these models resulted in a poor fit. This single-factor scoring method therefore may not be recommended based on our findings; its use could potentially hinder significant associations between FA and variables of interest, ultimately impacting on our understanding of FA in OCD. Accordingly, future research would benefit from the use of a 12-item FAS-PR total score and the two derived subscales instead to gain more meaningful insight into FA in OCD.

A second aim of the study was to compare the nature and extent of FA between mothers and fathers. The results confirm previous findings that parents commonly accommodate their child's OCD symptoms; the study however further extends previous literature by exploring both maternal and paternal accommodation and demonstrating that *both* engage high levels of accommodation on a *daily* basis. Furthermore, parents engage in similar patterns of accommodation; that is, both mothers and fathers reported provision of reassurance and participation in rituals as the two most frequent types of accommodation provided. Notably, significantly high correlation between mothers' and fathers' FAS scores ($r=.74$, $p<.01$) was also observed; as such, if one parent accommodates a child's symptoms, the other seems more likely to accommodate as well. Taken together, these findings suggest that parents take a unified approach to their child's OCD. Whilst both parents give accounts of being similarly drawn into rituals, however, parents differed in the extent of FA, with mothers accommodating more than fathers. This might simply relate to the amount of time a caregiver spends with a child with OCD, and the propensity to accommodate symptoms may be independent of the sex of the caregiver. We did not incorporate a measure of hours per day spent with the child and future studies may consider this. However, although mothers report more frequent accommodation, both parents remain highly accommodating of their child's symptoms, highlighting the importance of considering *both* parents in OCD assessment and treatment wherever possible to ensure that they are able to withdraw successfully from OCD symptoms, rather than having one parent inadvertently maintain a cycle of rituals and avoidance. To date, only two studies have examined whether relatives vary in their accommodation of OCD symptoms. Futh et al (2012) compared FA in 27 parent pairs and found no differences between mothers and fathers in their understanding, narrative, coping, and distress associated with parental accommodation. Gomes et al. (2014) on the other hand examined FA across different types of relatives (i.e. spouses, siblings, mothers, fathers, cousins), including mothers and fathers. Although fathers only made up a small proportion of their sample (n=7, 6%), no differences were noted between mothers and fathers in FA. The authors only found differences between spouses/partners and other family member, with the former engaging in significantly higher FA. Findings that mothers and fathers report a highly similar pattern of accommodation are therefore in line with these two previous investigations (Allsopp et al., 1990; Futh et al., 2012; Gomes et al., 2014). Unlike Futh and colleagues (2012) and Gomes et al (2014), however, we also found differences in the extent of FA by parent type, with

mothers accommodating their child's OCD symptoms to a far greater extent than fathers. These inconsistencies may be attributed to the relatively small number of fathers and lack of statistical power to identify significant parental differences in FAS in previous studies. Differences in sample characteristics, including symptom severity, may also partly account for these inconsistencies. For instance, Futh et al (2012) recruited self-reported OCD participants, for whom diagnoses were not confirmed; the sample in the current study on the other hand consisted of severe and complex OCD patients referred to a national specialist OCD Clinic. This is an important difference considering the confirmed association between symptom severity and FA (Storch et al., 2007, Stewart et al., 2008, Peris et al 2008, Merlo et al., 2009, Flessner et al., 2011, Lebowitz et al., 2014, Caporino et al., 2012, Futh et al., 2012).

This study sought to understand and compare the predictors of parental accommodation for mothers and fathers. The similarity of the findings across both mothers and fathers with regard to the predictors of accommodation is also noteworthy. Indeed, both maternal and paternal accommodating behaviours were predicted by child's OCD symptom severity and emotional and behavioural difficulties, and parental distress. The only difference in predictors was found in relation to paternal distress, in that fathers' DASS scores significantly predicted maternal accommodation of OCD symptoms, though the reverse did not hold true. Finding that OCD symptom severity predicted both maternal and paternal involvement in rituals is not surprising; in fact, this result is consistently emerging as one of the factors most relevant to understanding FA (Storch et al., 2007; Peris et al., 2008). Findings that symptom severity exerts an influence on mothers' as much as on factors' likelihood to accommodate, however, strongly supports the need for education regarding this coercive cycle and management strategies for such behaviour within the context of child OCD treatment. Such an approach should make sure to explore and address both maternal as well as paternal accommodation. As hypothesized, findings herein confirm a relationship between children's emotional and behavioural difficulties (as measured using the SDQ) as well as parental distress (as measured by the DASS) with FA of OCD symptoms; this was found to be equally true for mothers as well as father. In addition, fathers' distress predicted mothers FA. In a recent path analysis, Caporino et al (2012) found child's heightened emotional (specifically internalizing) difficulties as a pathway through which parental anxiety increased the risk for FA. Although the study clustered together data from both mothers and fathers, the field

would benefit from examining whether the mechanism of action applies in the same way to mothers as much as to fathers. Furthermore, the causal direction of this association cannot be established from our findings. Distressed parents could be more likely to accommodate in response to their child's emotional or coping difficulties. The reverse pattern however could also be true; that is, accommodating rituals could lead to increased parental distress. These two alternatives would have different implications for treatment and remain an important question to be addressed in future research. Based on their predictive effect, however, the results highlight the potential value of screening and providing additional support for children who present with generalized heightened emotional/behavioural difficulties and whose parents present with elevated distress levels. This could be achieved for instance by incorporating treatment components that directly address child as well as parental emotional difficulties in view of having a beneficial effect on reducing FA. The findings also reinforce the need to consider fathers' perspective as potentially indirectly influencing maternal accommodation.

Clinical practice guidelines currently recommend involvement of parents according to the needs of the child (NICE, 2005). Certainly there is a growing literature demonstrating good clinical outcomes for family-based CBT, with emerging evidence supporting the additive effect of parental involvement in OCD treatment (e.g. Storch et al., 2007; Freeman et al., 2014). One of the hypotheses of the current study was that both parental accommodation of symptoms would be associated with treatment response. The results were in support of this prediction. Indeed, when considered separately, both mothers and fathers accommodation significantly predicted post-treatment OCD severity (i.e. post-treatment CYBOCS scores) when controlling for baseline OCD severity, as previous research has shown (e.g. Merlo et al., 2010). Surprisingly however, when examining treatment response, defined as a significant (≥35%) reduction in CYBOCS scores from baseline to end of treatment, only paternal accommodation was found to predict treatment response, whilst maternal accommodation did not. When examining the subscales, results indicated that it was fathers' involvement in OCD rituals specifically to significantly predict treatment response. Of note, fathers' accommodation of their child's OCD symptoms predicted treatment response independently of whether mothers' levels of engagement in FA. Taken together, these results illustrate that both mothers' and fathers' accommodation impact on OCD severity post-treatment, consistently with the CBT theory of OCD for which FA impedes belief

discomfirmation and habituation, in turn maintaining symptoms. However, when examining treatment response, that is, whether accommodation predicted a *significant* reduction in symptoms, then fathers' involvement in rituals seems to play a crucial role. The large majority of studies to date have examined the association between FA and treatment outcomes using OCD symptom severity at post-treatment as the main outcome variable. Whilst this assures enough power for statistical purposes, the present study extended the current literature by having a large enough sample to examine the association of FA with treatment response, and clarifying the role of mothers' versus fathers' accommodation of OCD symptoms in CBT treatment outcomes for the first time. This is a novel finding that encourages more consideration and research on fathers' perspective in paediatric OCD. Whilst common clinical practice tends to privilege the participation of mothers in CBT treatment (Iversen et al., 2012), our results provide preliminary evidence to support fathers' involvement in their child's OCD treatment. For instance, research has shown that the involvement of fathers in treatment of disruptive behaviours in adolescence can lead to improved treatment outcomes (e.g. Bagner & Eyberg, 2013; Lundahl et al., 2008). In contrast to the literature on externalising behaviorus (Iversen et al., 2012), this issue remains to be examined in anxiety disorders and OCD in particular. The current findings however point to the potential benefit of examining this issue further and perhaps investigating the contribution of the involvement of fathers in treatment.

5. CLINICAL IMPLICATIONS AND FUTURE DIRECTIONS

The results have a number of clinical implications for the assessment and treatment of paediatric OCD and for future research.

Given its prevalence and association with treatment, routine screening of the nature and extent of FA by *both* parents is essential. As mothers are typically the primary caregiver involved in the young person's treatment, these findings support the need to consider fathers' response to their child OCD symptoms at assessment. To this end, the present study points to the potential value of using a 12-item, two subscales FAS-PR in future investigations on FA, rather than a 9- or 13-items FAS-PR total score, in order to assess FA in OCD. On a related note, the vast majority of existing studies on FA in OCD rely on the use of parent-report rating of accommodation with parents acting as the sole informants. This assessment method is subject to parents' insight, recall, and errors in self-observations. Future research may benefit from broadening the assessment of FA, by including clinician-administered measures and direct observations to support current findings.

Consistent with previous research, child's symptom severity and emotions/behavioural difficulties as well as parent distress levels were significant predictors of both maternal and paternal FA. Clinicians working with OCD should be aware of factors likely to predispose parents to FA and consider screening for these factors at assessment. To this end, self-report questionnaires, such as the FAS (Cavalcoressi et al., 1995), SDQ (Goodman, 1997; Goodman, 2001) and DASS (Lovibond et al., 1995), could be easily incorporated in clinical routine practice to screen for FA, child and parental distress levels at the start of treatment. These baseline measures could be used as clinical indicators for further assessment and to support family intervention.

In terms of implications for treatment, results underline the importance of educating both parents on the detrimental consequences of FA and addressing FA during treatment. A recent meta-analysis of FA in OCD indicated a significant medium effects size of the relationship between FA and OCD symptom severity (r=.35, 95% CI .23-.47) (Strauss et al., 2015). Theoretically, these findings lend some support to the idea of potential benefits of including family members in the OCD treatment; that is, outcomes may be improved when relatives are included in CBT sessions.

Emerging, yet conflicting, results have been reported in the literature, research on family-based CBT for OCD is emerging to examine the effectiveness of CBT treatment that addresses FA in OCD (e.g. Grunes et al., 2001; Reynolds et al., 2013). Indeed, in a small trial (n=28), greater OCD reductions were observed for patients assigned to a behaviour therapy plus family intervention group, compared to those in the behaviour therapy-only arm (Grunes et al., 2001). Furthermore, family members in the former group reported significantly less anxiety and depression post-treatment, with a potential impact on further reducing FA. These findings are however in contrast with those by Reynolds et al (2013). No significant differences were in fact observed in post-treatment OCD symptom severity when comparing outcomes for young people randomised to a course of individual CBT (n=25) versus parental enhanced CBT (n=25). Nonetheless, youths in the latter group reported greater reductions in anxiety symptoms; albeit in need of replication, the authors suggested that involvement of parents in their child's OCD treatment might result in more opportunities for generalisation of CBT principles and strategies for anxiety. Although FA is receiving more attention in recent years as a potential target for improving treatment outcomes for OCD, the current literature remains limited by small samples and most studies only involving mothers or clustering together different types of relatives. Future research is warranted to examine family-based CBT for OCD in larger samples as well as to examine the additional benefit of involving both parents, as opposed to one relative, in the family-based interventions for paediatric OCD. Moreover, causality between FA and treatment response cannot be established from this study. More research is therefore needed to confirm the direction of causation in order to inform intervention strategies; this could be addressed in randomized controlled trials which are beginning to emerge (e.g. Grunes et al., 2001; Freeman et al., 2014). Finally, longitudinal studies are needed to examine the link between FA and relapse in support of interventions targeting relapse prevention.

6. LIMITATIONS

A number of shortcomings ought to be considered when interpreting the current findings.

Whilst data on ethnicity and family composition were not available for all participants, anecdotally the sample consisted of largely white, British families; our findings therefore may not be generalizable to a range of families from differing ethnic backgrounds. Indeed, parenting styles have shown to differ across ethnic groups and cultures (Luis et al., 2008); it remains unclear therefore whether our results would apply to other ethnic groups. Recruitment of more culturally diverse OCD samples would help clarify this issue. In addition, the sample in this study consisted of severe and complex OCD patients referred to a national specialist OCD Clinic and may therefore not be entirely representative of paediatric OCD in the general population. Notwithstanding, the study offers unique information pertaining to how both mothers and fathers respond to a child with OCD.

As mentioned above, the study relied on parents as central informants, as opposed to using a more objective or clinician-administered measure of parental accommodation. Although the FAS-PR is a valid and reliable measure, findings are in need of replication using observational and clinician-administered measures of FA. Moreover, having tested the FAS-PR factor structure in a paediatric OCD sample, our results may not generalise to older OCD populations. Finally with regards to methodological limitations of the study, we did not include a measure of hours parents spent with their child; it is possible that the difference in the extent of FA between mothers and fathers is simply related to the time spent with their child as opposed to the sex of the caregiver; this requires investigation in future studies.

7. CONCLUSIONS

The present study supports the use of 12-items, 2 subscales FAS-PR as having the strongest factor analytic support to assess FA in paediatric OCD. Using this scale, we found that mothers and fathers are more similar in their propensity to accommodate child OCD than previous thought; results however also indicate differences in the extent of FA, with mothers engaging in more accommodation than fathers. Symptom severity, child emotional/behavioural difficulties, and parent distress were predictive of the risk for both parents to engage in accommodation of OCD

symptoms. Finally, the study confirms the association between maternal and paternal FA and post-treatment OCD severity; fathers' involvement in OCD rituals however played a crucial role in predicting a significant treatment response. These findings have important implication for clinical practice, highlighting the value of appropriate screening and targeting of both maternal and paternal symptom accommodation in paediatric OCD.

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9. APPENDIX

Appendix 1. Family Accommodation Scale- Parent Report (FAS-PR)

This questionnaire is for each PARENT/GUARDIAN to fill in.
Please state who has completed this form:
.....

Family Accommodation

Family participation in OCD behaviours during the past month		Never	1-3 times	1 or 2 times/ week	3-6 time/ week	Daily
1	How often did you reassure your child?	0	1	2	3	4
2	How often did you provide items for your child's compulsions?	0	1	2	3	4
3	How often did you participate in behaviours related to your child's compulsions?	0	1	2	3	4
4	How often did you assist your child in avoiding things that might make him/her more anxious?	0	1	2	3	4
5	Have you avoided doing things, going places, or being with people because of the your child's obsessive-compulsive disorder?	0	1	2	3	4
		No/ Never	Mild	Moderate	Severe	Extreme
6	Have you modified your family routine because of your child's symptoms?	0	1	2	3	4
7	Have you had to do some things for the family that are usually your child's responsibility?	0	1	2	3	4
8	Have you modified your work schedule because of your child's needs?	0	1	2	3	4
9	Have you modified your leisure activities because of your child's needs?	0	1	2	3	4
10	Does helping your child in these ways cause you distress?	0	1	2	3	4
11	Has your child become distressed/anxious when you have not provided assistance? If yes, to what degree?	0	1	2	3	4
12	Has your child become angry/abusive when you have not provided assistance? If yes, to what degree?	0	1	2	3	4
13	Has your child spent more time completing rituals when you have not provided assistance? If yes, how much more?	0	1	2	3	4

Transformation obsessions in paediatric obsessive-compulsive disorder: clinical presentation and outcomes of Cognitive Behaviour Therapy

SERVICE EVALUATION PROJECT

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1. ABSTRACT

Background: Transformation obsessions denote an under-reported symptom of Obsessive Compulsive Disorder (OCD), characterised by an excessive fear of turning into another person/object or acquiring unwanted characteristics. Relative to other OCD symptoms, little is known about the clinical presentation of transformation obsessions. In the adult literature, such obsessions are formulated as a contamination fear that is less responsive to exposure-based cognitive behaviour therapy (CBT) than other OCD symptoms.

Objective: The present audit aims to examine the clinical correlates and treatment prognosis of transformation obsessions.

Method: The sample consisted of 346 youths with a primary diagnosis of OCD. Patients with and without transformation obsessions were compared in terms of demographic and clinical characteristics, and CBT outcomes.

Results: 10% of the sample endorsed transformation obsessions. Patients with transformation obsessions were more likely to be boys, to be on augmented medication regimes, and to present with more severe obsessions at assessment. A factor analysis identified four major OCD symptom clusters, with transformation obsessions loading on a 'forbidden thoughts' factor alongside aggressive, sexual, and religious obsessions. No group differences in treatment outcomes were observed.

Conclusions: the audit provides the first empirical evidence on similarities and differences between paediatric OCD patients presenting with and without transformation obsessions. The findings suggest that transformation obsessions are best conceptualised as related to 'forbidden' obsessions and respond to exposure-based CBT as other OCD symptoms.

Service implications: recommendations for the N&S OCD Clinic include refinement of screening tools to address transformation obsessions and systematic screening at initial assessment. Dissemination of findings by the OCD Clinic, for instance through training and workshops, may help raise awareness of these less recognized OCD symptoms among non-specialist CAMHS and other relevant services to ensure adequate diagnosis and treatment.

2. INTRODUCTION

2.1. Paedriatric Obsessive Compulsive Disorder

Obsessive-Compulsive Disorder (OCD) is a psychiatric condition characterized by the presence of obsessions and/or compulsions (American Psychiatric Association, 2013). Obsessions are defined as recurrent and persistent thoughts, urges, or images that are experienced as intrusive, unwanted, and distressing for the sufferer. Compulsions on the other hand are repetitive behaviors or mental acts that the affected person performs in an attempt to prevent a feared event and/or reduce anxiety. A diagnosis of OCD is warranted if the obsessions or compulsions are time-consuming or cause clinically significant distress and/or functional impairment (American Psychiatric Association, 2013). Finally, a diagnosis of OCD can only be made once other conditions, notably substance use or general medical conditions, and alternative psychiatric disorders have been excluded (American Psychiatric Association, 2013).

Lifetime prevalence estimates for pediatric OCD have been found to range from 1 to 3% in the general population (Rasmussen & Eisen, 1998; Valleni-Basile et al., 1994), with a reported mean age of onset of around 10 years of age (Flament et al., 1990; Thomsen & Mikkelsen, 1991). While studies in adult OCD have shown an equal gender distribution, paediatric OCD samples have shown a different pattern, with male to female ratios of 2-3:1 in pre-pubertal onset OCD and 1:1.35 in post-pubertal OCD onset (Leonard et al., 1992). The onset and course of the disorder is usually gradual and can be chronic if untreated, often with waxing and waning of symptoms (Ravizza, Maina, & Bogetto, 1997; Skoog & Skoog, 1999, Mataix-Cols, Rauch, et al., 2002; Stewart et al., 2004). Although 70% of adults and children with OCD will meet criteria for one or more other comorbid psychiatric conditions (Fireman, Koran, Leventhal, & Jacobson, 2001), research has shown the most common one in pediatric OCD to be ADHD, followed by major depressive disorder, tic disorders, and oppositional defiant disorder (Leonard et al., 2001).

Finally, according to current evidence (e.g., Eddy, Dutra, Bradley, & Westen, 2004; Foa et al., 2005; Gava et al., 2007; O'Kearney, Anstey, & von Sanden, 2006; Soomro, Altman, Rajagopal, & Oakley-Browne, 2008; Pediatric OCD Treatment Study (POTS) Team, 2004) and recommendation by international and national treatment guidelines (American Psychiatric Association, 2007; National Institute for Health and Clinical Excellence, 2005), Cognitive-Behavior Therapy (CBT),

involving Exposure and Response Prevention (E/RP), and serotonin reuptake inhibitors (SRIs) are the first-line treatment for both adult and pediatric OCD.

2.2. Transformation obsessions

OCD encompasses a wide range of symptoms (Mataix-Cols *et al.*, 2005, Mataix-Cols *et al.*, 2008), including bizarre and magical obsessions. Volz and Heyman (2007) coined the term 'transformation obsessions' to refer to a subgroup of young people presenting with an excessive 'fear of turning into someone else or another object or acquiring unwanted characteristics'. Young people with transformation obsessions may, for example, have obsessional worries about becoming unpopular or losing their athletic skills or intelligence. Transformation obsessions can also manifest as a fear of turning into a specific person (e.g. Hitler) or even an animal (e.g. a rat; Volz & Heyman, 2007). This type of obsessional fear has also been recognised in the adult literature and referred to as 'morphing obsessions' (Rachman, 2006). Despite recognition of transformation or morphing obsessions as a symptom of OCD across the lifespan, relatively little is known about the clinical profile of this symptom.

In their description of transformation obsessions, Volz and Heyman (2007) described 9 young people aged 11-17 years presenting with this symptom seen in the National Specialist OCD clinic. Transformation obsessions were noted to be relatively rare, affecting only 9 out of 259 of referred young people with OCD. Importantly, the case series highlighted the common difficulty that clinicians experience in correctly diagnosing transformation obsessions due to their bizarre and unusual nature. In particular, misdiagnosis of transformation obsessions as being part of a psychotic disorder is a concern among this group.

In the adult OCD literature, transformation/morphing obsessions have been conceptualised as a form of 'mental' contamination, that is, feelings of dirtiness that are evoked in the absence of direct contact with a contaminant (Rachman, 2006; Warnock-Parkes et al., 2012) and associated with washing and cleaning rituals (Coughtrey et al., 2012). The association between transformation obsessions and contamination however remains to be confirmed. Indeed, there is no empirical evidence to support the notion that morphing/transformation obsessions fall within the contamination dimension of OCD. To clarify the conceptualization and nature of

transformation obsessions, their relation to OCD symptom dimensions requires further investigation.

In the adult literature, a few case studies on adult OCD have suggested that mental contamination, including transformation obsessions, may be more treatment-resistant than other symptom dimensions of OCD, and may require a modified treatment approach (e.g. extended or adapted protocols). For example, Warnock-Parkes *et al.* (2012) described a case of a man with a 20 year history of mental contamination associated with traumatic memories who had not responded to E/RP-based CBT delivered through a specialist OCD service. He received modified cognitive therapy incorporating imagery work to address his appraisals of key events that had given rise to his feelings of contamination. Following the course of cognitive therapy, his symptoms decreased from the severe to sub-clinical range. This case report highlights the need for further research to test the extent to which cognitive therapy, using techniques such as imagery rescripting, is superior to E/RP-based CBT in the treatment of mental contamination. With regard to transformation/morphing obsessions specifically, it remains to be confirmed whether CBT requires modification to successfully address these obsessions. Whilst Volz and Heyman (2007) recommend standard E/RP-based CBT for the young people in their case series, no study has yet explored whether transformation obsessions respond to CBT to the same extent as other OCD symptoms in young people.

3. AIMS AND HYPOTHESES

To summarise, OCD is a heterogeneous condition encompassing a wide range of symptoms. Transformation obsessions refer to a 'fear of turning into someone else or another object or acquiring unwanted characteristics'. Little is known about the clinical presentation and nature of transformation obsessions. Whilst NICE guidelines for paediatric OCD recommend E/RP-based CBT as the first-line treatment, studies on adult OCD samples have suggested that these obsessions may be more treatment-resistant than other symptoms of OCD and may require a modified treatment approach.

The OCD and Related Disorders Clinic has developed a standardised CBT protocol for OCD, and previous audits have demonstrated that this treatment protocol is effective for the majority of individuals treated at the clinic. To date, however, it is unclear whether this treatment protocol is appropriate for individuals presenting with 'transformation obsessions'.

The aim of the present audit is to examine and compare clinical features and CBT outcomes for OCD patients with and without transformation obsessions; this audit will help identify issues that need to be addressed when working with the specified group of young people and therefore highlight potential areas for possible improvement in service provision.

In light of the above, the objectives of the present service evaluation project are as follows:

1) To identify the rate of pediatric OCD patients seen at the National Specialist OCD service endorsing transformation obsessions
2) To examine the clinical presentation of OCD patients endorsing transformation obsessions in comparison to those with other forms of OCD
3) To assess how this cohort of patients responded to the standardized CBT protocol used the OCD & Related Disorders Clinic, in comparisons to young people with other OCD symptoms.

The audit addressed these objectives using routinely collected baseline and end-of-treatment data. Examination of the nature and impact of transformation obsessions on treatment and service provision is important to ensure the needs of these users are being met.

4. METHODS

The audit was approved by the CAMHS (Child and Adolescent Mental Health Services) Audit committee at South London and Maudsley NHS Foundation Trust.

4.1. National Specialist OCD & Related Disorders Service

This audit took place at the National Specialist OCD & Related Disorders service at the Maudsley Hospital, a Tier 4 outpatient National and Specialist CAMHS service. The OCD Clinic consists of a team of mental health professionals (mainly psychologists and psychiatrists), offering assessment and treatment for young people up to 18 years of age with OCD and related disorders.

All patients who are seen in the service undergo a thorough multidisciplinary assessment process, involving structured clinical interviews with the child and their parents in order to obtain information on OCD symptomotology and severity and a detailed account on the developmental and family history. Diagnoses are confirmed by the specialist multidisciplinary team at the end of this comprehensive assessment.

A per NICE guidelines, the first line psychological treatment offered by the Clinic is CBT, incorporating psycho-education, E/RP, and relapse prevention. The intervention is delivered according to an established treatment protocol, developed by the clinic (Turner, 2008), with previous audits supporting its clinical utility and effectiveness for the majority of individuals treated at the clinic. In addition to CBT, psychotropic medication is offered and monitored by the service.

4.2. Participants

A total of 346 young people who were referred to the National and Specialist Pediatric OCD & Related Disorders Clinic at the Maudsley Hospital (London) and who met ICD-10 diagnostic criteria for OCD were the subjects of this audit. A total of 217 OCD patients received CBT treatment at the clinic; of these, a proportion (31.3%) also received SSRI medication. In most cases medication had reached a stable dose before CBT commenced. Those receiving medication were more likely to be slightly older ($p=0.02$) and present with more severe OCD ($p<0.01$).

4.3. Measures

The *Children's Yale-Brown Obsessive-Compulsive Scale* (CY-BOCS) (Scahill *et al.*, 1997) is the gold standard clinic-administered measure of OCD severity. It includes a symptom checklist followed

by 10 items assessing the severity of obsessions and compulsions in terms of time, interference, distress, resistance and control. Total OCD severity score ranges from 0-40. The CY-BOCS demonstrated excellent reliability and validity properties (Storch et al., 2004).

The clinic routinely conducts a CY-BOCS interview at the initial assessment and at the end of treatment. Data from CY-BOCS assessment interviews was used to identify OCD patients endorsing 'transformation obsessions'. For confirmation and clarification, information on transformation obsessions was also elicited from clinicians who conducted assessment and treatment sessions.

The *Children's Obsessive-Compulsive Inventory* (ChOCI) (Shafran *et al.*, 2003) is a questionnaire assessing obsessive-compulsive symptoms in young people and has a parent and child version. The ChOCI has shown good internal consistency and criterion validity and strongly correlates with the CY-BOCS (Shafran *et al.*, 2003, Uher *et al.*, 2008).

The *Strengths and Difficulties Questionnaire* (SDQ) (Goodman, 2001) is a 25-item questionnaire capturing emotional, conduct, hyperactivity/inattention, peer problems, and pro-social behaviour, including questions assessing impact. The measure has a parent and child version, is widely used across a range of clinical settings, and has been shown to have good psychometric properties (Goodman, 2001).

The *Beck Depression Inventory for Youth* (BDI-Y*)* (Beck JS *et al.,* 2001) is a widely-used 20 item self-report measure for depressive symptoms, which has good internal consistency and test-criterion validity (Beck JS *et al.,* 2001).

The *Family Accommodation Scale* (FAS) (Calvocoressi *et al.*, 1995, Calvocoressi *et al.*, 1999) is a parent-report measure of parental involvement in their child's OCD symptoms. It consists of 4 subscales (Participation, Modification, Distress and Consequences), and has been shown to have excellent internal consistency and good convergent and criterion validity (Pinto *et al.*, 2013).

The *Children's Global Assessment Scale* (CGAS) is a valid and reliable clinician-administered measure of overall functioning, on a 0–100 point scale (Bird *et al.*, 1987, Shaffer *et al.*, 1983).

4.4. Statistical analyses

Data were analyzed using SPSS version 21.0 for Windows. Between-group differences were tested using Chi-square tests for categorical data, Mann-Whitney U-test for ordinal or non-normally distributed data, and Student's t tests for continuous data.

To explore the relationship of transformation obsessions to other OCD symptom dimensions, a Principal Component Analysis (PCA) with varimax rotation was carried out using the number of endorsed symptoms under each major symptom category of the CY-BOCS (Mataix-Cols et al., 2005).

Finally, a mixed model Analysis of Variance (ANOVA) was used to test for a differential effect of group (transformation obsessions / no transformation obsessions) on responsiveness to treatment. All statistical tests were two-tailed. Significance level was set at $p < .05$.

5. RESULTS

OCD patients with and without transformation obsessions were compared on key demographic and clinical variables as well as on response to CBT. The audit also attempted to clarify the relation of transformation obsessions to other OCD symptoms using factor analysis. Below a summary of the main findings.

5.1. Sample characteristics

The sample consisted of 189 (54.6%) boys and 157 (45.5%) girls, with a mean age of 14.4 years (SD 2.2; range 7-18); age at onset of OCD was 10.7 years (SD 3.1). The mean total CY-BOCS score at assessment was 26.4 (SD 5.6), indicative of moderate OCD severity.

5.2. Rate, clinical presentation, and conceptualisation of transformation obsessions

A total of 35 (10.1%) OCD patients endorsed transformation obsessions. Demographic and clinical characteristics of OCD patients with and without transformation obsessions are presented in *Table 2*.

Individuals with transformation obsessions were more likely to be male, as indicated by a significant group difference in gender ($\chi^2 = 6.07$, df = 1, p=0.014). No differences emerged with regard to age at assessment, years until first treatment contact, and past psychological and pharmacological treatments (all p>0.05). Patients with transformation obsessions, however, were significantly more likely to be on augmented medication than individuals without such obsessions ($\chi^2 = 7.42$, df = 1, p=0.006).

With respect to OCD symptom severity, the two groups did not differ on the CY-BOCS total score (p=0.11) nor on the compulsions subscale (p=0.45). There were however significant differences on the CY-BOCS obsessions subscale (p=0.01). The group with transformation obsessions reported spending more time worrying and experiencing greater interference and a lack of control over obsessions than those without transformation obsessions.

No statistically significant differences were found with respect to child and parent self-report measures of OCD, depression, emotional and behavioural difficulties, global functioning, and family accommodation (*Table 2*).

The PCA identified four factors explaining 51.3% of the total variance (*Table 3*). The first factor ('*Contamination*') explained 14.3% of the variance and included contamination and somatic obsessions, as well as cleaning and checking compulsions. The second ('*Hoarding*') and third

('*Symmetry*') factors explained 12.8% and 12.4% of the variance, respectively. Saving obsessions, hoarding and ordering/arranging compulsions loaded on the second factor, while the third factor included symmetry obsessions and repeating, counting, and ordering compulsions. Finally, transformation obsessions loaded on a fourth factor ('*Forbidden Thoughts*'; explaining 11.8% of the variance), alongside aggressive, sexual, and religious obsessions.

Table 2. Demographic and clinical characteristics of patients with and without transformation obsessions

Variable	Participants with transformation obsessions (N=35, 10.1%)		Participants without transformation obsessions (N=311, 89.9%)		Statistics	
DEMOGRAPHICS	*N*	*%*	*N*	*%*	*Chi-square*	*P*
Males	26	74.3	163	52.4	6.07	0.014*
On SRI medication for OCD	18	51.4	109	35.0	3.63	0.057
On SRI plus augmentation	7	20.0	21	6.8	7.42	0.006*
Previous CBT for OCD	18	52.9	119	39.8	2.17	0.140
Previous other therapy	4	12.5	47	18.3	0.656	0.418
	Mean	*SD*	*Mean*	*SD*	*Student's t*	*P*
Age at assessment (yr)	14.71	1.90	14.33	2.25	-0.931	0.352
Age of OCD onset (yr)	11.03	3.02	10.69	3.14	-0.661	0.509
Years till first treatment	1.32	1.82	1.95	2.47	-1.448	0.148
OCD MEASURES						
CY-BOCS Total	27.49	5.38	26.28	5.62	-1.557	0.119
Obsessions Subscale	13.97	2.71	12.78	2.99	-2.537	0.011*
Compulsions Subscale	13.51	13.0	3.41	2.92	-0.753	0.451
ChOCI Child-report	29.25	7.28	29.57	8.83	1.253	0.819
ChOCI Parent-report	34.28	9.17	31.36	9.47	0.212	0.144
FAS Mother	30.00	12.96	24.98	13.99	0.781	0.094
OTHER CLINICAL MEASURES						
BDI-Y (T score)	63.33	12.54	59.65	12.52	-1.451	0.147
SDQ Child	20.40	4.05	19.69	4.99	0.549	0.592
SDQ Parent	19.47	3.68	19.43	5.11	2.274	0.974
CGAS	43.03	7.68	46.87	10.92	1.919	0.056

Abbreviations: OCD, Obsessive Compulsive Disorder; SRI, selective re-uptake inhibitors; CY-BOCS, Children Yale-Brown Obsessive Compulsive Scale; ChOCI, Children's Obsessive-Compulsive Inventory; BDI-Y, Beck Depression Inventory; SDQ, Strengths and Difficulties Questionnaire; CGAS, Children' Global Assessment Scale.

Table 3. Factor structure of OCD symptom dimensions (N=346)

	Factor Loadings				
	Contamination	*Hoarding*	*Symmetry*	*Forbidden Thoughts*	*Total*
Obsessions					
aggressive	.196	.322	.074	**.697**	
contamination	**.846**	.044	-.115	.085	
sexual	.034	-.090	.001	**.631**	
hoarding/saving	.075	**.815**	-.042	.061	
symmetry	.053	-.049	**.740**	.095	
somatic	**.479**	.171	.055	.142	
religious	.130	.051	.154	**.648**	
Transformation	-.332	-.038	.019	**.453**	
Compulsions					
cleaning	**.786**	-.069	-.052	-.136	
checking	**.495**	.333	.204	.197	
repeating	-.094	.017	**.765**	.095	
counting	-.006	.368	**.568**	.101	
ordering/arranging	.075	**.433**	.422	-.171	
hoarding	.104	**.722**	.113	.039	
% of variance	14.3	12.7	12.4	11.8	51.3

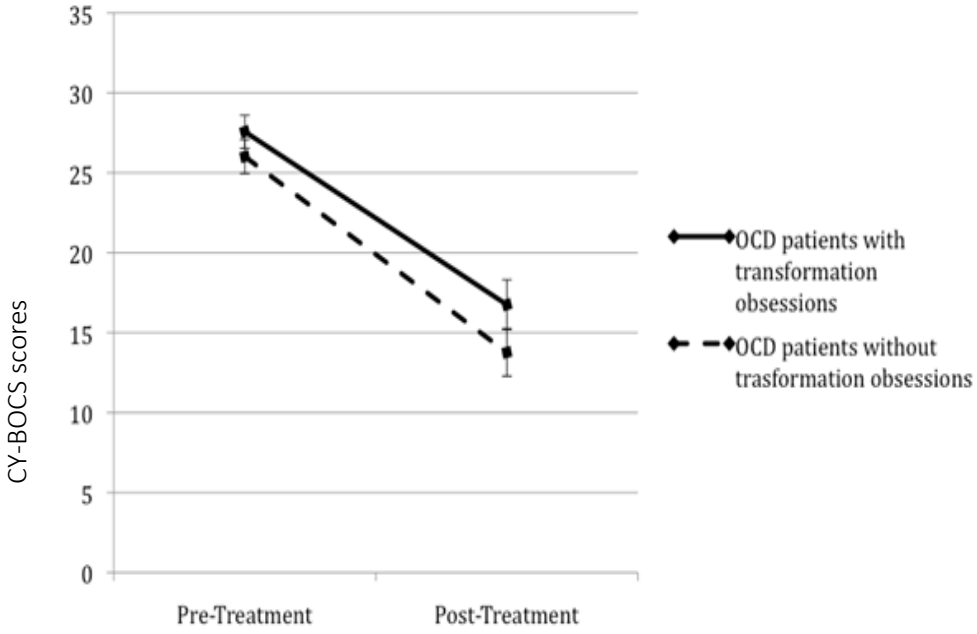
Note: highest loadings highlighted in bold.

5.3. Treatment outcomes

A mixed-model ANOVA with time as the within-subject factor (pre- versus post-treatment) and group (with versus without transformation obsessions) as the between-subjects factor revealed a main effect of time [F(1, 214)= 228.582, p <0.001], but no Time x Group interaction [F(1, 214)=0.858, p=0.355], indicating that both patient groups responded equally well to CBT treatment (*Figure 1*). Similar patterns of findings were observed for the obsessions and compulsions CY-BOCS subscales. For obsessions, there was a main effect of time [F(1, 212)=229.252, p<0.001] and no Time x Group interaction [F(1,212)=0.002, p=0.966]; and for

compulsions, again there was a Main Effect of Time [F(1, 212)=183.005, p<0.001], but no Time x Group interaction [F(1,212)=19.096, p=0.169].

Figure 1. Treatment outcomes for paediatric OCD patients with transformation obsessions (N=28) and without transformation obsession (N=188)



Abbreviations: CY-BOCS, Children Yale-Brown Obsessive Compulsive Scale.

6. DISCUSSION

This project was designed to investigate the rate, clinical presentation, conceptualisation, and CBT outcomes for youths presenting with transformation obsessions. Findings will be discussed in relation to each aim.

6.1. Rate of transformation obsessions

Results indicate that transformation obsessions were relatively common in our clinical sample, with approximately 10% of youth with a primary diagnosis of OCD endorsing fears of turning into someone else or another object or acquiring unwanted characteristics. This is higher than the prevalence rate of approximately 3% that was previously reported by Volz & Heyman (2007) in the same clinic, and may reflect an increased awareness and more careful screening of transformation obsessions before referral and/or within the specialist OCD clinic. This finding on its own supports the need for routine screening as this symptom may be not be uncommon as anticipated.

6.2. Clinical presentation and conceptualisation of transformation obsessions

The second aim was to examine the clinical presentation of young people endorsing transformation obsessions and to clarify the relation of transformation obsessions to OCD symptom dimensions.

Findings suggest that transformation obsessions are comparable to other forms of OCD with regard to phenomenology; indeed, similarly to other forms of OCD symptoms, patients with transformation obsessions endorse worries and compulsions that are time consuming, distressing, interfering, and hard to resist and control.

Differences were also noted; as such, transformation obsessions were common among boys and associated with greater severity on the obsessions subscale on the CY-BOCS. Among other differences, the group of patients with transformation obsessions were also more likely to be on augmented medication than those without such symptoms. The reasons for increased use of augmented medication is unclear and whilst we cannot make firm conclusions, the finding raises the question as to whether these symptoms are associated with a poor or incomplete response to Selective Serotonin Reuptake Inhibitors (SSRIs). Alternatively, it may also be plausible that clinicians are more likely to think of alternative pharmacological management plans in response to the bizarre nature or the distress associated with these obsessions. For example, as described

by Volz & Heyman (2007), clinicians misinterpret transformation obsessions as an indication of an emerging psychosis, and may therefore be inclined to add an anti-psychotic medication. Should this be the case, raising awareness of transformation obsessions among clinicians is important to avoid diagnostic confusion and delayed or erroneous treatment.

Findings from the current study also have important implications for clarifying the relationship of transformation obsessions to OCD symptom dimensions. In the adult literature, morphing/transformation obsessions are conceptualised as a form of 'mental contamination', falling within the contamination dimension of OCD. A factor analysis identified a four-factor OCD symptom structure, that is largely comparable to previous investigations (Bloch *et al.*, 2008, Mataix-Cols *et al.*, 2008). In contrast to suggestions from the adult literature, transformation obsessions were found to load on a 'forbidden thoughts' dimension alongside aggressive, sexual, and religious obsessions. This finding is consistent with Volz & Heyman (2007), where only two out of the nine young people with transformation obsessions presented with washing or cleaning compulsions; the finding also supports patients' description of their obsessions as a fear of harm coming to them. Whilst tentative, overall our results encourage the need for routine screening of these symptoms in OCD, particularly among patients reporting 'forbidden obsessions'.

As no currently available OCD measures specifically addresses items relating to transformation obsessions, it would be paramount as a next step to refine screening procedures and include items in standard OCD checklists that elicit such obsessions. For instance, clinicians might ask 'do you ever worry that you may turn into someone or something else or that you may take on some unwanted/negative characteristics?' A systematic screening approach would result in early identification and intervention tailored to address these more obscure OCD symptoms.

6.3. Treatment outcomes

The third and final aim of this audit was to evaluate treatment outcomes for young people with transformation obsessions compared to those without such obsessions.

Results from the current audit indicate that young people with transformation obsessions respond equally well to the standard E/RP-based CBT protocol, as compared to patients with other obsessions. The clinic data suggest that this treatment protocol is appropriate for individuals presenting with 'transformation obsessions' and does not require a modified treatment approach (e.g. extended or adapted protocols).

Taken together, these findings suggest that transformation obsessions in paediatric OCD are not as rare a phenomenon as was previously thought and that, whilst somewhat unusual in content, they exhibit similar characteristics to other OCD symptoms and should therefore be formulated and treated as any other obsession. The audit indicates that this cohort positively responded to the ERP-based CBT protocol delivered by the specialist OCD service.

The audit also does not support the association between transformation obsession and contamination that has been anecdotally reported in the adult literature, and cautiously note the association between these obsessions and 'forbidden obsessions' for clinicians.

7. LIMITATIONS

A number of limitations of the present study should be considered. First, whilst the study reports on the frequency of transformation obsessions among children and adolescents with a primary OCD diagnosis, replications of these findings are needed; indeed, whether the occurrence of these obsessions is even higher remains to be properly examined utilizing prospective screening. Second, some of the participants were taking SSRI medication while receiving CBT. While in most cases medication was at a stable dose before CBT commenced, some patients were prescribed new medications or their doses changed during the CBT, according to clinical needs. We therefore cannot infer the effects of CBT alone on outcomes. Finally, any subsequent studies on transformation obsessions may benefit from inclusion of more comprehensive assessments tools and from follow-up data to assess treatment outcomes in more depth.

8. IMPLICATIONS AND SERVICE RECOMMENDATIONS

The audit has highlighted a number of important issues and recommendations to be considered by the clinic. These are summarised as action points for the clinic below:

- Refinement of screening tools to incorporate items addressing transformation obsessions. This may be achieved through insertion of an item on transformation obsessions on the 'OCD obsessions checklist' of the Child Yale-Brown Obsessive Compulsive Disorder Scale (C-YBOCS) that is administered by clinicians at the clinic at initial assessment and on self-report measures of OCD (e.g. Obsessive-Compulsive Inventory).
- Systematic assessment and screening of transformation obsessions within the clinic, particularly when assessing and/or treating boys and those presenting with 'forbidden obsessions'
- Disseminating findings to non-specialist CAMHS and/or other appropriate services involved in the treatment of pediatric OCD in order to raise awareness of these less recognized OCD symptoms, including transformation obsessions, and therefore to ensure adequate diagnoses and interventions. Strategies for dissemination include training and workshops (incorporating case presentation, skills demonstrations, group discussion and feedback), supervision and telephone consultations.

9. DISSEMINATION OF FINDINGS

The results of this audit and recommendations were presented to the OCD team's research 'away day' on 1st May 2014. The team was interested and pleased to hear about the outcomes, which led to a discussion about implications for the service. As reported in the methods, the OCD team conducts thorough clinical diagnostic interviews at the initial assessment, which includes the administration of an OCD symptom checklist followed by the CY-BOCS to assess the person's OCD symptom severity. Given the high staff turnover, to avoid inadvertently failing to enquire on these symptoms, the team agreed to add an item to the checklist to directly probe and elicit transformation obsessions at intake. This may help identify and address these obsessions sooner in the treatment process; using a prospective approach to the identification of TO may also help to gather a better estimate of the true occurrence of these symptoms within an OCD service.

As part of the dissemination process, the audit was submitted and accepted for publication in *Journal of Behavior Therapy and Experimental Psychiatry*. Furthermore, the N&S OCD Team offers extensive programme of training in the assessment, treatment and management of different aspects of OCD through national seminars and workshops; dissemination of the project results will occur also through these seminars and workshops.

10. SERVICE USER INVOLVEMENT

Service users have been involved in planning and execution of the audit, as described here below. Service users (sufferers, carers, and mental health professionals) were consulted on the clinical utility of the present audit. A young service user wrote:

> *"I think research could be helpful. Although I have felt able to talk about my obsessions, they were not labelled as transformation obsessions until I came for specialist treatment. As soon as I knew what they were I felt much more able to tackle my difficulties. It was like a light bulb moment when it all made sense. It's really scary when you're fighting something that's unknown so it helped so much to know and understand what was going on. If transformation obsessions could be recognised even sooner that can only be a good thing."*

In exploring the usefulness of understanding transformation obsessions and their impact on OCD treatment, a mental health professional reported:

> *"Patients who present with these obsessions may present a real challenge clinically, so more research to inform and guide diagnosis and treatment would be really helpful."*

The results of this service evaluation were fed back to the OCD clinic; implementation of the recommendations are aimed to improve the service delivery and the experience of youths presenting with transformation obsessions.

11. LEADERSHIP

The Leadership Framework Model (http://www.leadershipacademy.nhs.uk/discover/leadership-framework/), developed by the NHS Leadership Academy, comprises of 7 domains for demonstrating effective leadership skills and behaviours: demonstrating personal qualities, working with others, managing services, improving services, setting direction, creating the vision, delivering the strategies. Below is a description and examples of how the present audit allowed opportunities for development of leadership skills.

In terms of 'personal qualities', the audit was approached in an organized and methodical way. An interest in this area of investigation was expressed to the team from the outset, which resulted in the opportunity to take a leading role in the development and progress of the project.

'Working with others' was the foundation of this audit, and observable through the collaborative approach during all stages of the audit, from conceptualisation of the topic to be examined, data collection, the writing process, to dissemination of the findings. Recommendations were openly discussed; the audit encouraged sharing of perspectives and feedback. Strategies for the dissemination of the findings were designed jointly with the team with a view of improving service provision (e.g. through refinement of screening tools). The audit allowed development of skills within the domain 'improving services'; this was achieved through facilitation of discussion (e.g. presentation and publication of findings) on recommendations for the service and action points to improve the service provided.

12. SUMMARY AND CONCLUSIONS

Whilst associated with certain clinical features (in terms of gender, medication, and severity of obsessions), overall transformation obsessions do not differ significantly on demographic features and prognosis, compared with other forms of obsessional thoughts. The study validates the effectiveness of using the Clinic's CBT protocols to successfully address the core fear underlying transformation obsessions. Given the occurrence of these symptoms in paediatric OCD and their potential functional impairment, clinicians should refine screening tools systematically assess for transformation obsessions. Improved screening may encourage early detection and appropriate treatment, potentially helping minimize the long-term risk associated with any OCD symptomatology.

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